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An unusual presentation of synovial chondromatosis of the knee in a 10-year-old girl

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ABSTRACT

Synovial chondromatosis commonly occurs in the anterior compartment of the knee joint, predominantly in middle-aged men. It is relatively unusual in female children and is rarely encountered in the synovium beneath the meniscus. The present report describes a rare case of synovial chondromatosis that developed in the synovium just inferior to both menisci of the right knee in a 10-year-old girl. At this unusual age and location, there is a greater probability of missed diagnosis, due to the lack of definite informative incidence, and difficulty in finding the lesions during arthroscopic examinations. In the present case, multiple loose bodies were hidden by the meniscus, and thus, there were no structural abnormalities in the initial arthroscopic views before probing the meniscus. After careful inspection, we found numerous cartilaginous loose bodies and removed them as much as possible with arthroscopy. © 2017 Turkish Association of Orthopaedics and Traumatology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/ 4.0/).

Introduction

Synovial chondromatosis is an unusual proliferative and metaplastic disorder characterized by the formation of multiple cartilaginous nodules in the synovial membrane of the joint, tendon sheath, and bursa.¹ It usually occurs in middle-aged and elderly patients with a minor male predominance.^{2,3} Although synovial chondromatosis can develop in any joint where synovium exists, it is most prevalent in the knee followed by the hip joint.⁴ Within the knee, the disease predominantly occurs in the anterior compartment such as the suprapatellar pouch, medial or lateral gutter, and infrapatellar fat pad.⁵ Recently, there have been some reports regarding synovial chondromatosis in the unusual location of the knee joints, such as posterior compartment.^{6,7} However, to our knowledge, there have been no reported cases occurred in the submeniscal area of the knee joint.

The present report describes a case of a 10-year-old girl who presented with synovial chondromatosis that had occurred in the synovium just inferior to both menisci in the right knee, with a

E-mail addresses: dricarus@naver.com (H.J. Cho), jwjd714@naver.com (J.D. Suh). Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology. locking sensation for 6 months. In the present case, synovial chondromatosis occurred in the common joint but in a less common age group and in an unusual location of the joint. In the arthroscopic examination, multiple loose bodies were hidden by the meniscus initially. After careful inspection with probing, multiple loose bodies were successfully managed with arthroscopic removal.

Case report

A 10-year-old girl presented to our orthopaedic outpatient clinic with a chief complaint of flexion deformity of the right knee joint and limping gait that had begun a half year previously without any specific injury event. Physical examination revealed lack of extension by approximately 20°, but knee flexion was unimpaired. Tenderness over the medial and lateral joint line was also noted. No effusion was visible, and there was no palpable mass across the joint line. During the gait, a slight limp was observed due to the fixed flexion deformity of the right knee. Laboratory test results were within normal limits, and there was no evidence for infectious or other inflammatory arthritis. There was also no specific familial history for any musculoskeletal disorders.

Plain radiographs revealed no mass or bony lesion in the right knee joint. Magnetic resonance images (MRI) revealed innumerable tiny nodular lesions in anterior and posterior part of the knee just inferior to the both menisci (Fig. 1). These lesions could only be

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Fig. 1. Magnetic resonance imaging. T2 axial (A) and PD sagittal (B) MRI showed innumerable low signal intensity, tiny, nodular lesions (arrowhead) in the anterior and posterior parts of the knee just inferior to both menisci. T1 sagittal (C) MRI, which were cut with the same slice cut as in Fig. 1B, showed obscure synovial lesions without mass configuration.



Fig. 2. Arthroscopic view. Initial arthroscopic view (A) of the medial compartment through the anterolateral portal showed normal configuration of the articular cartilage and meniscus. Elevation of medial (B) and lateral (C, D) menisci reveled multiple cartilaginous loose bodies.

identified as numerous small round masses with low signal intensity on T2-weighted axial and proton density (PD) sagittal images. However, on T1-weighted sequence, the lesions appeared isointense to the skeletal muscle, and thus could not be distinguished from the surrounding tissues. Other intra-articular structures, including cartilage, meniscus, and ligaments, showed normal appearance. Surgery was decided to resolve the symptoms and to make a definite diagnosis.

The patient was placed in the supine position, and surgery was performed under tourniquet control. Diagnostic arthroscopy was performed using a 30° arthroscope through an anterolateral portal. On the arthroscopic examination, there were no floating loose bodies in the knee joints. Meniscus, cartilage and ligament structures were normal in appearance. However, elevation of both menisci by the probe revealed numerous tiny cartilaginous materials, which were loosely attached in the synovium just inferior to the meniscus (Fig. 2). The loose bodies were removed and washed out as much as possible, and synovial tissue was debrided in the parts where there were cartilaginous bodies in it. We then created anteromedial and anterolateral submeniscal portals to access loose bodies located inferior to the anterior horn of both menisci. Via the submeniscal portal, numerous chondromatous loose bodies were also removed (Fig. 3). Histologic examination revealed a neoplastic

cartilaginous tumor resembling hyaline cartilage with a lobulated configuration encapsulated by synovial tissue (Fig. 4). In the lacunae of the cartilaginous tissue, there were clusters of chondrocytes with single nuclei of regular contour and diameter, which was consistent with a diagnosis of synovial chondromatosis.



Fig. 3. Gross photograph. There were numerous brilliant white, cartilage-like loose bodies each with an average diameter of 2–5 mm.



Fig. 4. Histological examination of a retrieved loose body. Note that the body was composed of hyaline cartilage which was located beneath the thin synovial layer (arrow). Hyaline cartilage was filled interiorly with clusters of chondrocyte (hematoxylin and eosin stain, \times 40).

After the operation, the symptoms improved dramatically. Flexion contracture of the knee joint and limping gait were spontaneously resolved. Since the lesions were not detected preoperatively using the simple radiographs, serial MRI follow-up was recommended for the detection of recurrence. However, in our case, the patient's parent refused it due to the economic burden. Nevertheless, at 12- months after surgery, the patient showed no signs of recurrent disease.

Discussion

Intra-articular synovial chondromatosis most commonly involves the knee joint.⁴ The condition can be classified as generalized or localized. The localized form can theoretically be present in any compartment of the knee joint that contains synovium, but it usually develops in the anterior compartment.⁵ If the lesions develop in these common locations of the knee joint, they can be easily detected in arthroscopy. The term "snow storm knee" has been used to describe this condition as seen in arthroscopy, which presents typical appearances of radiopaque calcification and ossification on the synovial membrane in the involved joint.¹ However, if the lesions develop in an unusual location, this typical arthroscopic appearance may not be present. Furthermore, if the lesions are hidden by other intra-articular structure, they may be difficult to find, and may be misdiagnosed as normal. In the present case, there were no floating materials and no structural abnormalities in the initial arthroscopic views before probing the meniscus. After careful inspection, we found the numerous cartilaginous loose bodies, which were hidden by the meniscus in the submeniscal area.

Clinically, synovial chondromatosis can look similar to other proliferative disease processes of the knee synovium, such as chronic synovial proliferation, pigmented villonodular synovitis and multiple rice bodies. The diagnostic similarities between these intra-articular pathologies are particularly significant in cases of synovial chondromatosis without calcified or ossified bodies.^{8,9} Prior studies have demonstrated that the classic finding of x-ray calcifications may be absent in 5–30% of cases of synovial chondromatosis.⁴ In cases without demonstrable findings of calcification or ossification, MRI has been shown to be the preferred imaging modality, rather than X-ray or computed

chontomographic arthrography, to evaluate synovial dromatosis.^{8,10} Typical MRI appearance of synovial chondromatosis was characterized as lobulated, homogeneous, intermediate, intra-articular signal intensity similar to that of muscle on T1-weighted images, with high signal intensity on T2weighted images and focal areas of low signal intensity with all pulse sequences.¹⁰ Pigmented villonodular synovitis, in contrast, typically demonstrates low-intensity proliferative synovial changes on T1-and T2-weighted images due to synovial iron contents.¹¹ Multiple rice bodies appear as non-attenuating homogenous soft tissue nodules that are isointense in the T1weighted sequences and minimally hyperintense when compared with muscle in the T2-weighted series.¹² In the present case, the multiple loose bodies appeared isointense to the skeletal muscle on T1-weighted images and were virtually unrecognizable. They could only be identified as numerous small round masses with subtle decreased signal intensity on T2-weighted axial and PD sagittal images at the meniscal level. These MRI findings in the present case were not compatible with the typical MRI appearance of synovial chondromatosis, and they were not sufficient to distinguish it from other synovial proliferative synovial disease. Therefore, we could not confirm the present case as synovial chondromatosis prior to histopathologic examination Although MRI reliably reveals the presence of intra-articular loose bodies, it may be non-specific, and difficult to differentiate from other intraarticular pathologies.

Synovial chondromatosis is a disease that principally affects the middle-aged and elderly population with a minor male predominance.^{2,3} It is relatively rare in female children. A search of peerreviewed literature revealed only two cases of synovial chondromatosis in female children.^{13,14} In those two reports, the number of loose bodies was less than three, and the size of them was quite large, which could be easily recognized on the preoperative MRI and arthroscopic examinations. Furthermore, the masses were located in relatively common locations, and thus they could be easily removed on the arthroscopy. However, in the present cases, the loose bodies were numerous and extremely small. MRI findings of the present case were non-specific. In the arthroscopic examination, because the loose bodies were located inferior to both menisci, we could not easily identify them before probing the meniscus.

In conclusion, synovial chondromatosis should always kept in mind as a rare but possible differential diagnosis, particularly if female children complain of a locked knee without a history of trauma. Although it commonly occurs in the anterior and posterior compartments of the knee, it can also develop in the synovium just beneath the meniscus. At this unusual location, there is generally a greater possibility of missed diagnosis and insufficient removal of loose bodies during the arthroscopic examination. Therefore, careful inspection for locating and removing loose bodies during the arthroscopy is crucial in the diagnosis and treatment of synovial chondromatosis in unusual locations.

Conflicts of interest

None of the authors have relevant conflicts of interest to declare.

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