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Case Report

Arthroscopic Treatment of Atypical Synovial Osteochondromatosis of Infrapatellar Fat Pad: A 4-Year Follow-up Case Report and Literature Review

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Abstract

Primary synovial osteochondromatosis, also known as synovial chondromatosis, is a rare benign arthropathy caused by synovial cartilage metaplasia. The tumor is rarely seen outside the knee joint cavity, especially in the infrapatellar fat pad (IFP). This case presents an older woman who complained of left knee joint pain with a growing isolated mass for 2 years. Physical examination showed that a hard mass was palpable below the patella. X-ray and computed tomography examination of the knee joint revealed cluster calcification foci under the patella. Under arthroscopy, the mass was found in the IFP and removed, with pathological examination indicating synovial osteochondromatosis. The patient recovered well after the operation, and no sign of recurrence was observed at the 4-year follow-up. This case report proposes that synovial osteochondromatosis in atypical sites, such as IFP, should heighten our attention and could be treated by arthroscopy.

Introduction

Synovial osteochondromatosis is a relatively rare disease in which the synovium develops metaplastic transformation and forms osteochondral bodies in the synovial cavity.[1] Large joint cavities, such as the knee, hip and elbow joint cavities, are particularly vulnerable, but no case of synovial osteochondromatosis located outside the joint cavity has been reported.[2] In this report, we have diagnosed a novel case of infrapatellar fat pad osteochondromatosis (IFPO), which originated from the synovium, was located in the IFP and was treated by knee arthroscopy.

Keywords: Synovial osteochondromatosis; Loose body; Infrapatellar fat pad; Knee; Arthroscopy.

Abbreviations: IFP, infrapatellar fat pad; IFPO, infrapatellar fat pad osteochondromatosis; CT, computed tomography; IKDC, International Knee Documentation Committee.

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Case Report

A 64-year-old woman presented a two-year history of mild pain in the anterior aspect of the left knee with a hard, slow-growing mass around the knee joint. She had no history of trauma, but her knees suffered repetitive minor injuries when kneeling on the ground to do farm work. Physical examination showed that there was a hard, unmovable mass, which could be palpated from the medial or lateral side of the patellar tendon in the infrapatellar region. There was a mild left knee effusion. The range of motion of the lesioned knee joint was 110°~20°~0°, which was 20° smaller than that of the contralateral knee. No sign of an unstable knee joint or inflammatory reaction was found. Routine laboratory examinations, including complete blood count, blood biochemical indexes, and urine analysis, were all within normal limits. Radiographs of the left knee showed an osseous mass within the IFP (Fig. 1a). Computed tomography (CT) showed trabeculated ossification in the IFP (Fig. 1b). Based on the results of physical examination and imaging, it was suspected that metaplastic bone was formed in the IFP. Fine needle aspiration for cytology failed, as the needle could not enter the bony mass.

Under arthroscopy, the tumor was found within the IFP without communication with the joint cavity or patella tendon (Fig. 2a). Numerous white pigeon-egg-sized loose bodies, also called "white stones," were observed outside the articular cavity of the left knee (Fig. 2b). General observation revealed that the mass was

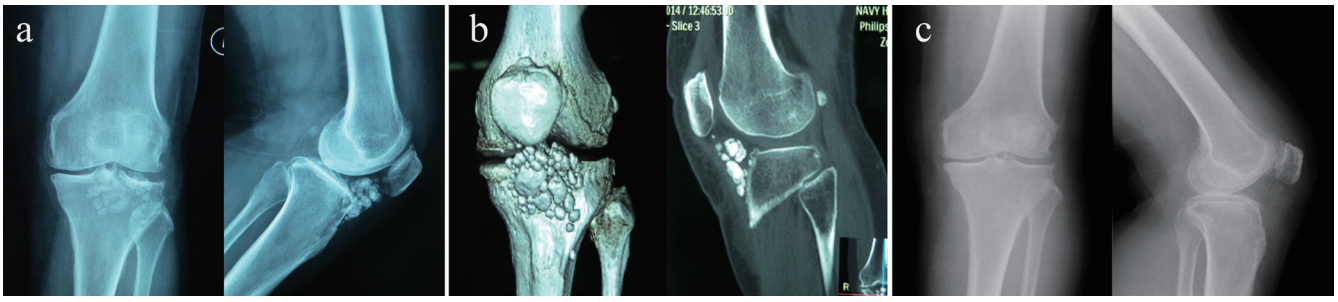

Fig. 1. Imaging examination of the patient before and after the operation. (a) X-ray image of anteroposterior and lateral view of the lesioned knee before the operation. (b) Computed tomography scanning of the lesioned knee. (c) Postoperative X-ray examination of the left knee, showing no loose body residue.

composed of cartilage and irregular nodule-like bones wrapped by fibrous membrane, with an average size of 1×2 cm and a maximum size of 4×3 cm (Fig. 2c). The synovium that proliferated around the IFP was removed as much as possible, and loose bodies were washed out. Through histological examination, no cell with nuclear atypia or mitosis was observed, and no obvious pathological change was found in the surrounding soft tissue (Fig. 3).

Postoperative X-ray examination of the left knee joint showed that there were no loose body residues in the knee joint. Within 24 h after the operation, an ice compress was suggested, and full weight bearing and movement without restriction on the ground was allowed. Three days after the operation, the knee joint pain was obviously relieved, the range of knee flexion and extension was increased to 0°~150°, and the overextension and flexion test became negative. According to the International Knee Documentation Committee (IKDC) standard, the score was increased from 72 before the operation to 93 points after the operation. Daily life was not affected, but occasional discomfort of the knee when it rains was a complaint. During the 4-year follow-up, no clinical or radiological sign of recurrence was observed.

Discussion

As the most common bone tumor, osteochondromas is considered to be a disease of proliferative-developmental abnormality of the growth plate rather than a real tumor.[1] Most patients develop osteochondromas in their fifties, and the incidence in male patients is 2-3 times higher than that in females.[3,4] The main clinical manifestations of the disease are joint pain, swelling, joint effusion, tenderness, and dyskinesia. Since only high-density tumors with complete calcification can be displayed via X-ray examination, CT or MRI examination is often needed. Trauma is usually found accompanied with osteochondromas and has been generally accepted as a possible inducement.[5] The exact cause of osteochondromas remains unclear, but chondrometaplasia and ossification of synovial mesenchymal cells have been suggested to be the primary cause. While some scholars believe that the pathological changes of synovial osteochondroma originate from the metaplasia of mesenchymal cells, one hypothesis proposes that the disease originates from the metaplasia of adipose tissue in lipoma.[6] According to the pathological stage, Milgram *et al.*[7] divided the disease into three stages: 1) active synovial lesion with hyaline cartilage metaplasia without calcification; 2) loose body in synovial tissue, without loose body falling off; and 3) loose body composed of cartilage tissue.[7] In this study, the patient was in stage 3. Loose bodies can also be present in patients with advanced osteoarthritis or exfoliative osteochondritis but are usually accompanied by narrowing of the joint space and proliferation of local osteophytes or by corresponding bone defects and mainly a single loose body. Osteochondromas are commonly found in the articular cavity, especially the knee joint, and are rare outside the cavity. Herein, we described a patient who presented with synovial osteochondromatosis in the IFP that developed from potential osteochondroma. It is probable that osteochondroma caused inflammatory changes within the IFP, which led to osteochondroma metaplasia and the development of synovial osteochondromatosis.[8]

In this case, the patient was exposed to minor injuries, owing to her occupation as a farmer. Repeated minor injuries to the IFP were probably associated with this tumor in this patient. After the operation, the patient followed our instructions of avoiding farm work

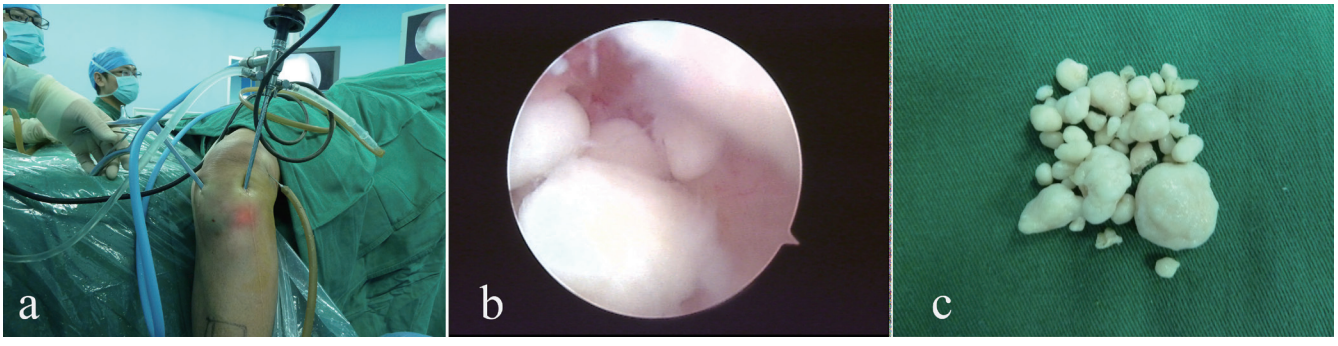

Fig. 2. The IFPO was treated by arthroscopy. (a) Loose bodies treated by arthroscopic treatment. (b) Under arthroscopy, the so-called "white stones" were located within the IFP, and no loose body was seen in the cavity of the knee joint. (c) Removal of different sizes of loose bodies via washing out. IFP, infrapatellar fat pad; IFPO, infrapatellar fat pad osteochondromatosis.

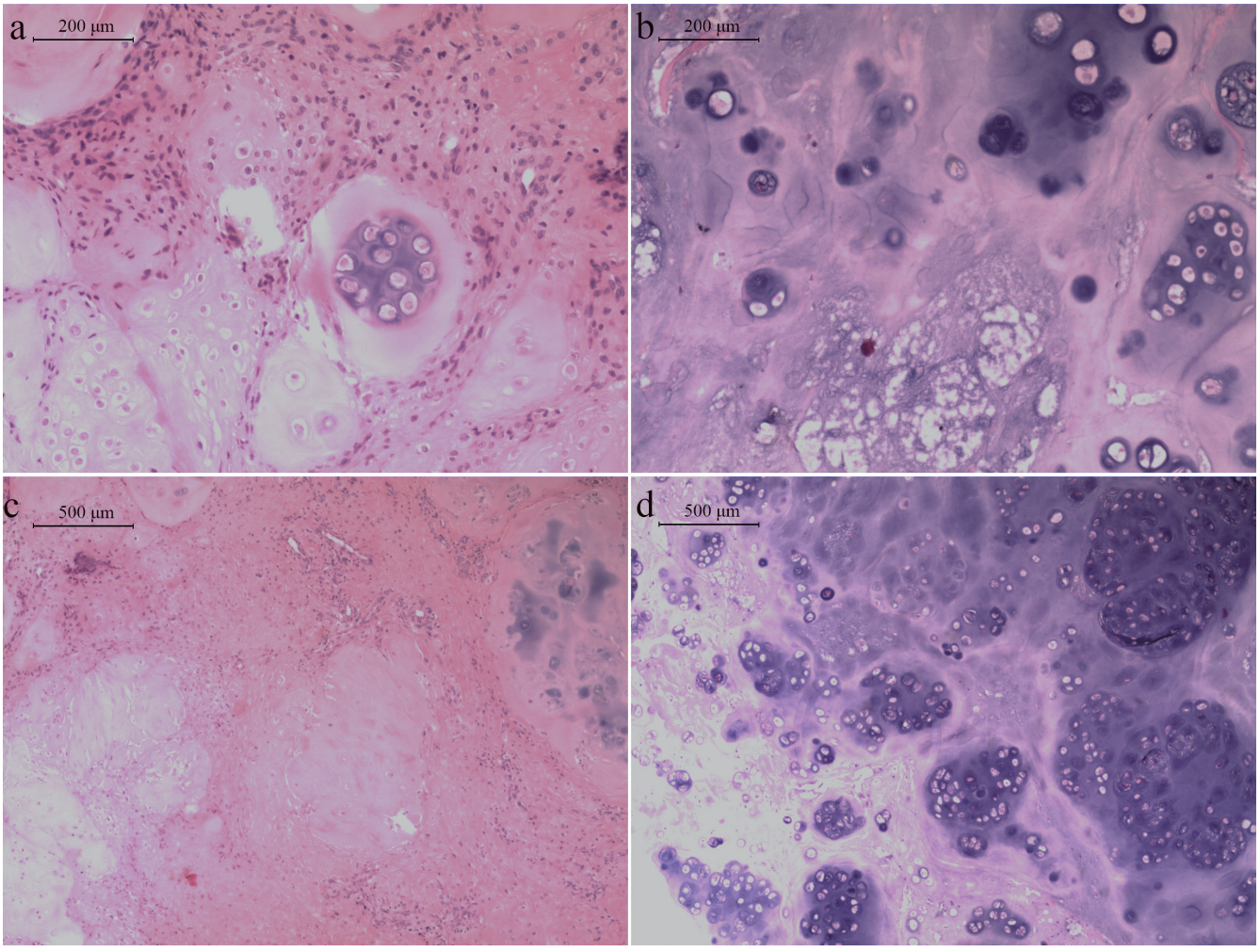

Fig. 3. Pathological examination showed that the loose bodies were synovial osteochondromatosis. (a, b) Magnification: ×100, scale bar = 200 μm. (c, d) Magnification: ×40, scale bar = 500 μm.

to prevent repeated minor injuries to the knee; there was no sign of recurrence during the 4-year follow-up. Concomitant degenerative arthritis is another clinical manifestation of the advanced stage of this disease. With the passage of time, primary synovial osteochondromatosis could lead to cartilage degeneration by increasing mechanical wear via loose bodies and nutrient deprivation to the articular cartilage.[9] Besides, degenerative arthritis could, in turn, lead to secondary synovial osteochondromatosis.[9] The existence of severe osteoarthritis also presents a challenge for orthopedic surgeons. Since radiotherapy and chemotherapy are not effective for synovial osteochondromatosis, surgical excision is generally recommended. As a minimally invasive operation, arthroscopic surgery enables early postoperative rehabilitation exercise and inhibits joint function damage. Other injuries in the joint can also be examined and treated under arthroscopy. Therefore, traditional incision surgery for synovial osteochondromatosis has gradually been replaced by arthroscopic minimally invasive treatment. In cases that involve local intra-articular lesions, complete removal of abnormal synovium is expected to provide a cure. On the other hand, systemic intra-articular diseases with pain and swelling require total synovectomy and loose bodies resection.

At present, the treatment of synovial osteochondromatosis is still controversial. It has been reported that arthroscopic removal of the loose bodies is sufficient, while synovectomy is unnecessary.[10] However, some scholars suggest that synovectomy should be performed at the same time.[11] Osti *et al*.[12] reported that some patients still relapsed after arthroscopic removal of loose body and partial synovectomy and then chose open surgery. Since the synovium is considered to be the origin of the disease, it is also recommended to remove as much synovium as possible. A systematic review showed that most IFPO patients were treated with a simple excision via an opening incision.[13] Since mechanical injuries may lead to degenerative arthritis and potentially to secondary synovial osteochondromatosis, arthroscopy treatment should be preferred to minimize the mechanical injuries caused by surgery and the economic burden of patients.

Conclusions

Atypical synovial osteochondromatosis can occur outside the joint cavity, such as IFP, for which minimally invasive knee arthroscopy is an effective treatment.

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Conflict of interest

CC has been an editorial board member of *Exploratory Research and Hypothesis in Medicine* since November 2020. The authors declare no other conflict of interests.

Author contributions

Study concept and design (BZB, CC), acquisition of data (BZB, ZHB), analysis and interpretation of data (BZB, ZC), drafting of the manuscript (ZC, CC), critical revision of the manuscript for important intellectual content (BZB, CC), administrative, technical, or material support, study supervision (BZB, ZC).

Ethical statement

All procedures performed in studies involving human participants were in accordance with the ethical standards of the Institutional and National Research Committee and with the 1964 Helsinki Declaration and its later amendments, or comparable ethical standards. The patient gave written informed consent in accordance with the Declaration of Helsinki. The authors obtained the written informed consent from the patient for publication of this case report and the accompanying images.

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