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## Case Report

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### Intra-Articular Osteochondroma of the Knee Joint in a Patient with Hereditary Multiple Osteochondromatosis

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**Abstract** Hereditary multiple osteochondromatosis (HMO) is characterized by multiple osteochondroma (OC) arising from enchondrally formed bones<sup>1)</sup>. Such lesions are seen most commonly in the forearm, knee and hip joint. In long bones, OC arises adjacent to the physis and tends to remain in the metaphyseal lesion<sup>2)</sup>. Therefore, it is rare that OC is located intra-articular of the joint and only few cases of intra-articular OC have been previously reported<sup>3)~5)</sup>. Intra-articular OC could appear as a single loose body in a joint cavity<sup>6)</sup>. Thus, other joint disorders, such as osteochondritis dissecans should be ruled out in such cases. We herein report a case with an intra-articular OC in a knee joint cavity in the patient with HMO.

**Key words** : Hereditary multiple osteochondromatosis, intra-articular, knee

#### Case report

This report was approved by the patient. An 18-year-old man presented with a click in his left knee of 2 year's duration. He also noticed bumps around both knees. His mother and sister had been diagnosed to have HMO. He had no history of knee trauma or knee surgery. A physical examination revealed a hard bony mass without any tenderness on the medial side of both knees. He had tenderness on the medial joint line of the left knee. Although he could walk without pain, a click was audible at the posterior lesion of the left knee at flexion to extension. The range of

motion of the left knee was 0 to 125 degrees and he felt a slight pain at full flexion. He had no symptoms in his right knee. Plain radiographs showed many sessile OCs at the distal metaphysis of the femur and proximal metaphysis of the tibia. In the left knee joint, we observed an ossified and round loose body at the site of an intra-condylar lesion (Fig. 1A and B). MRI without gadolinium application showed the mass to be located behind the posterior cruciate ligament (PCL) (Fig. 2A ; sagittal plane, 2B and C ; coronal plane). The intensity of the mass was largely hypointense on gradient echo (GRE) sequence (Fig. 2 A and B). We also found an exstosis at a medial cortex of the tibia (arrow) (Fig. 2B). Although the mass had no continuity with the femur, it was located very close to the intra-condylar lesion of the femur (arrow) (Fig. 2C).

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Since his knee pain had worsened and the range of motion of left knee had decreased (0 to 90 degrees), we carried out arthroscopy for both treatment purpose and to make a definite diagnosis. Our pre-operative diagnosis was the intra-articular loose body of unknown origin of the knee joint. An arthroscopic examination showed no pathological findings of articular cartilage and meniscus of the left knee. However, we found a 2 x 1 cm oval mass with a cartilage surface behind the PCL. The mass was not connected to its surroundings by bone bridges, thus indicating that the mass appeared as a loose body (Fig. 3). We found a slight synovitis in the affected knee. The mass was removed and then it was subjected to a histological examination. A pathological study revealed the mass to have mature trabecular bone with fatty marrow tissue covered with a cartilage cap (Fig. 4). This feature is compatible with OC and there was no evidence of malignancy. After arthroscopy, the click and pain in the left knee both disappeared and the range of motion thereafter recovered to 150 degrees. One year after removal, the knee was symptom free and no signs of recurrence were evident on radiograph.

### Discussion

In patients with HMO, OC usually occurs around the growth plate of long bone in childhood and then it moves toward diaphysis according to the growth<sup>1)</sup>. Therefore, it is rare that OC is located intra-articular of the joint. Osteochondroma may become symptomatic if it irritates soft tissue, such as the tendon, muscle, and nerve<sup>7)</sup>. In the present case, the patient felt a click and pain in the affected knee. OC was located in the posterior cavity of the knee joint as a loose body. We found only

one previous report of intra-articular OC of the knee joints in a patient with HMO<sup>8)</sup>. However, in that case, the OC showed typical pedunculated shape and diagnosis was not complicated. In contrast, the OC in our case appeared as an osteochondral loose body within the knee joint and a definitive diagnosis of this case was more challenging than that of the former case.

Preoperative imaging including radiographs and MRI showed a completely intra-articular mass in the posterior joint cavity. The lesion appeared to be a single mass of cartilagenous or osteocartilagenous tissue, which had grown into the posterior cavity of the knee joint, thus grossly resembling a single loose body. An arthroscopic examination showed the mass to be completely intra-articular; no communication with the bone was found. Such lesions should be differentiated from loose bodies resulting from other causes including osteochondritis dissecans, detached osteophyte, and osteochondral fractures. However, the patient had no history of trauma to the left knee. Furthermore, we also observed no defect in the articular cartilage in the left knee joint. We therefore could rule out the possibilities of those diseases.

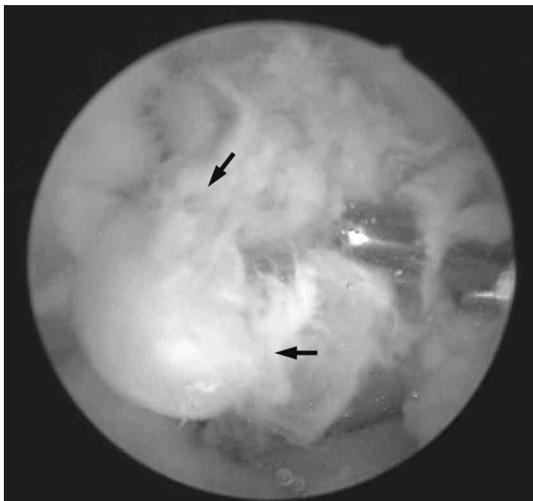
Synovial osteochondromatosis (SOC) is an uncommon disorder characterized by the formation of multiple cartilagenous nodules within the synovium. These nodules are often covered by a fine fibrous layer and synovial lining cells<sup>8)</sup>. In the usual form of SOC, multiple lobules of proliferation cartilage in the synovial tissue of joint are present. However, in certain instance in SOC, focal intrasynovial osteochondral nodules may form, thus bearing some resemblance to intra-articular OC<sup>9)</sup>. In our case, a histological examination of the loose body demonstrated the typical features of an OC.



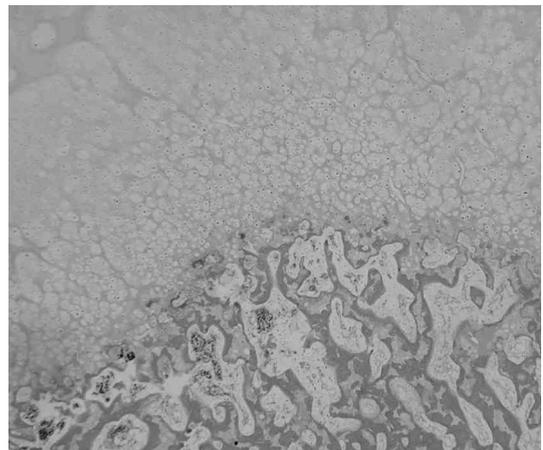
**Fig. 1** Radiographs of the left knee (A ; lateral view, B ; anteroposterior view) demonstrating an ossified and round mass in the intra-condylar lesion of the posterior cavity of the left knee (arrows). Note the many sessile osteochondromas at the distal metaphysis of the femur and the proximal metaphysis of the tibia (arrow heads in B).



**Fig. 2** MRI of the left knee showed the mass behind the posterior cruciate ligament (PCL). (arrows) (A ; sagittal view, B, C ; coronal view). The intensity of the mass was largely hypointense at gradient echo sequence (TR 490 msec, TE 16 msec, FA 16.0). We also found an exostosis at a medial cortex of the tibia (arrow in B). Although the mass was located very closely to the intra-condylar lesion of the femur, it had no continuity with the femur (arrow in C).



**Fig. 3** An arthroscopic view of the intra-condylar lesion of the left knee. The arrows indicate osteochondroma.



**Fig. 4** A low power photomicrograph of osteochondroma. The lesions showing the mature trabecular bone covered with a cartilage cap. H&E, original magnification x 40.

The lesion had a cartilage cap and secondary bone formation through a process similar to normal enchondral ossification. Only small areas of synovial lining cells were observed, thus indicating that a diagnosis of SOC was unlikely. In addition, the size of the loose body was relatively large (2 x 1cm), while the size of an individual loose body found in case of SOC was rarely greater than 1 cm in diameter<sup>10)</sup>. Taken together, we diagnosed the loose body to be intra-articular OC.

The origin and pathogenesis of the presented intra-articular OC were another interesting issue. We hypothesized two mechanisms. First, we speculated that intra-articular OC was actually a secondary disorder caused by shedding of the pedunculated OC in the joint. If OC originates at the posterior end plate of tibia or femur, it could protrude into a joint cavity. Then it might be torn and the OC thereafter truly became loose within the joint. In line with this idea regarding our case, we found a bony protrusion at the intra-condylar lesion of the femur just adjacent to the OC. The protrusion might be a remnant of a pedicle of the pedunculated OC (Fig. 2C, arrow). On the other hand, OC could arise in the juxta-articular soft tissue without any attachment of bone, thus indicating that OC may originate in the extraskeletal tissue. Extraskeletal OC is a rare finding<sup>11)</sup>. Up to now, few cases of extraskeletal OC in and around knee joints have been reported<sup>4)6)10)12)</sup>. However, in this case, we have no evidences, which support the mass to be extraskeletal OC.

Recently, one case of an arthroscopic resection of intra-articular of OC in the knee joint has been reported<sup>3)</sup>. An arthroscopic resection of the OC relieved the symptoms in that case. In our case, an arthros-

copic resection of the OC also eliminated the click and pain of the patient, thus further supporting the usefulness of arthroscopy for the treatment of the intra-articular OC. In conclusion, OC could appear as intra-articular loose body which might thus cause a progressive limitation of the joint function. A surgical removal, including an arthroscopic resection, should therefore be considered for both treatment purpose and for making a definitive diagnosis.

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(和文抄録)

## 遺伝性多発性外骨腫症に伴う膝関節内外骨腫の1例

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遺伝性多発性外骨腫症は、前腕、膝、股関節周囲などに多発する骨軟骨腫を特徴とする疾患である。多くの症例で、骨軟骨腫は長管骨の骨幹端近傍に存在し、関節内発生例の報告は少ない。今回、骨軟骨腫を膝関節内に発生した遺伝性多発性外骨腫症の希な1例を経験したため報告する。