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## Large para-articular osteochondroma of the knee joint: a case report

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A unique case of large intra-articular osteochondroma of the knee of a 15-year duration is presented along with a review of the literature. The tumor may have remained asymptomatic for such a long period because of its slow growth and stretch elongation of the quadriceps mechanism.

**Key words:** Intra-articular osteochondroma; knee; para-articular osteochondroma.

Osteochondroma is a common benign neoplasm that develops mostly around the growth plate of long bones, especially the knee. The tumor demonstrates mature bone with a cartilaginous cap and continuity with the medullary cavity, and usually ceases growth with closure of the growth plate. Infrequently, this neoplasm arises in the soft tissues. Extraskeletal osteochondromas are rare and their biological behavior is not well-known. [1] More than 80% of these tumors occur in the hands and feet. [2-4] Para-articular osteochondroma compromises a small subgroup of these extraskeletal osteochondroma that commonly affect the knee joint, with a few involving the hip, ankle and elbow. [5-7] The concept of ESPAOC (extraskeletal paraarticular osteochondroma) was first introduced in 1958 by Jaffe, who used the synonymous terms 'para-articular chondromas' and 'intracapsular chondromas' to describe osteochondral metaplasia occurring in the fibrous joint capsule or the soft tissue adjacent to the

joint.[8] However, it was Milgram and Dunn who first used the term 'para-articular osteochondroma' and differentiated these lesions from synovial osteochondromatosis. In joints with a large capsular space, such as the patella-femoral joint, osteochondroma can remain intracapsular.[9,10]

We report a patient with large extraskeletal intraarticular osteochondroma in the anterior portion of the knee joint and present a review of the literature regarding para-articular/intra-articular osteochondromas.

## Case report

A 52-year-old male presented with a fifteen year history of a slow-growing mass in the anterior portion of the left knee with slight restriction of movement (Fig. 1). There was no swelling on any other part of the body. The patient had no family history of tumors, metabolic or rheumatic conditions or prior history of trauma or infection. Physical examination revealed a

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Fig. 1. Clinical photograph showing mass in the infrapatellar area of the left knee. Swelling extends both on the medial and lateral side of the patellar ligament. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]



**Fig. 2.** Lateral plain X-rays showing a well-circumscribed solitary mass in the anterior aspect of the knee in the infrapatellar area.

hard non-tender mass on the anterior aspect of the knee below the patella which was palpable, both on the medial and lateral aspect of the patellar tendon. The patient had full extension and a flexion of 100°.



**Fig. 3.** MRI showing a well-demarcated inhomogeneous lesion which had low signal mostly on T1-weighted images.

Plain radiographs showed a well-delineated mass in the anterior aspect of the knee in the infrapatellar area (Fig. 2). Femoral and tibial condyles were normal. MRI showed a well-demarcated inhomogeneous lesion which had low signal on T1-weighted images and a mixed high and low signal on T2-weighted images (Fig. 3).

Routine hematological investigations were within normal limits. The decision for an excision biopsy was taken with a preoperative provisional diagnosis of benign osteochondral lesion.

A midline vertical incision was made with the patient under regional anesthesia and tourniquet control. The mass lied medially, posterior and lateral to the patellar tendon, almost completely replacing the infrapatellar fat pad. Patellar tendon was stretched over the mass (Fig. 4a). The mass mimicked the femoral condyles and the intercondylar notch (Fig. 4b) and the patellar tendon passed over the notch/groove. The excised sample was sent for laboratory testing and confirmation of benign osteochondroma was made. The mass was unable to be removed en bloc due to its large size and was instead divided into two halves with each half removed by retracting the patellar tendon to either side of the mass (Figs. 4b-d). The tibial plateau and patellar tendon were not involved (Fig. 4e). The

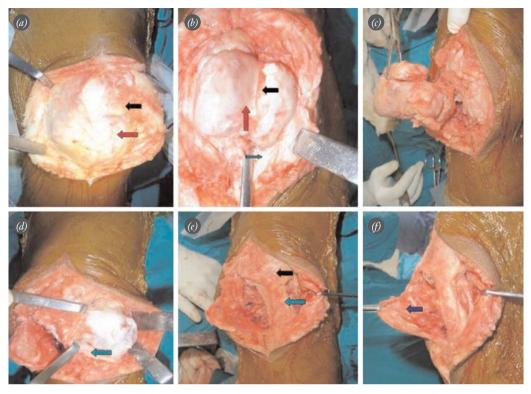


Fig. 4. Perioperative photographs: (a) Mass in the infrapatellar area extending on both sides of the patellar ligament (ligament marked by outlined red arrow; patella marked by black arrow). (b) Mass simulating femoral condyle and intercondylar notch in the center (outlined red arrow). Mass was divided into two halves for removal (black arrow). Patellar ligament was retracted laterally (outlined blue arrow below). (c) Medial half of the mass being removed. (d) Lateral half of the mass being removed. Patellar ligament was retracted medially (arrow). (e) Perioperative view post mass removal (patellar ligament marked with green arrow and patella with black arrow). (f) Photograph showing the lax capsule (arrow). [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

redundant part of the capsule was excised and capsular repair was carried out (Fig. 4f).

The resected specimen was 15.5x8x3.8 cm with smooth margins and a glistening surface. The medial portion of the mass showed a nodular appearance and a groove for the patellar tendon was located in the middle (Fig. 5).

On histopathological examination, the mass showed a well-formed, partly-lobulated mature hyaline cartilage surrounded by fibrous tissue at the periphery. The hyaline cartilage changed to mature trabecular bone in the center. Foci of active endochondral ossification were found at the interface between the mature bone and the well-differentiated cartilaginous cap. There was no evidence of malignant features or mitotic activity. There was no chondroblastic or chondrogenic differentiation or zonal phenomenon. No synovial tissue was identified (Fig. 6). The final pathological diagnosis of extra skeletal osteochondroma was established.

Postoperative recovery was uneventful. The patient returned to activities of daily living and regained full flexion two weeks after the procedure. The patient was asymptomatic at the final follow-up 26 months after the excision, with no clinical or radiographic signs of recurrence of the lesion.

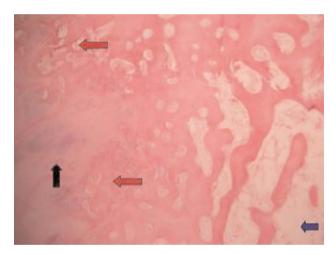
## Discussion

To our knowledge, 45 extraskeletal osteochondromas, not extending to the joint space, have been reported in the literature under different names, such as para-articular, soft tissue, capsular, intracapsular, or intra-articular osteochondromas, ossification of the infrapatellar fat pad, and ossifying chondromas. [1,5,11-22]

By far, the most common site of the 45 ESPAOCs in the knee joint is the infrapatellar fat pad, while a few cases have been described in the suprapatellar pouch and recently in the posterior aspect of the knee. [6,23,24] Bleshman and Levy reported an intra-articular osteochondroma of the hip with lateral displacement of the femoral head. [25]



**Fig. 5.** Excised tumor mass. Tumor was divided in the center for removal (medial half marked with red arrow, lateral half with black arrow, and the center with blue arrow). [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]



**Fig. 6.** Photomicrograph showing a cap of mature hyaline cartilage at the periphery (black arrow on the left) and endochondral ossification in the center. Mature bone trabeculae containing bone marrow are also seen (outlined red arrows) and fat cells of the infrapatellar fat pad are seen on right side of the photograph (blue arrow below). (H&E x200) Histopathology confirmed classical picture of osteochondroma. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

As early as 1941, Robillard reported a large osteo-cartilaginous mass in the infrapatellar area, which was labeled as ossification of infrapatellar fat pad. [22] Roth [26] reported an ossifying chondroma, while Kautz [12] found 4 cases which he labeled 'capsular osteoma of the knee joint'. Reith et al. stated that term 'capsular osteoma' was inappropriate as a cartilaginous element was present in these growths, and that para-articular chondroma was not valid as all cases had trabecular bone and predominant enchondral ossification. [1] They evolved

criterion based on these concepts to correctly identify these unique lesions; a single dominant mass, lesion consisting of bone and cartilage organized like conventional osteochondroma, and lesions arising purely from extra-articular non-synovial tissue.

The pathogenesis of such tumors remains controversial. Lesions appear de novo without any apparent precursor. [2] Metaplasia of pluripotent cell line derived from the joint synovium, tenosynovium, and connective tissue has been proposed as the origin of the tumor cells. [13] While typically occurring in adults, no age limit has been reported. Antecedent trauma has been reported in a few cases although the stimuli in most cases remain unknown. [27] The behavior of extraskeletal osteochondroma is poorly characterized but limited data suggests a benign make-up with rare local recurrence. [28]

Radiographic findings of extraskeletal osteochondroma typically consist of a well-circumscribed, lobulated mass with a dense central calcification or areas of ossification. (T demonstrates the extraskeletal location and central dense calcification or ossification of the osteochondroma. MRI shows a well-demarcated inhomogeneous lesion which has a mostly low signal on T1-weighted images and mixed high and low signals on T2-weighted images. Areas of mature ossification have intermediate T2 signal intensity with the exception of the densely calcified areas which have a low signal intensity. [4,29]

Various possible differential diagnoses, such as myositis ossificans, lipomatous lesion, tumoral calcinosis, extraskeletal chondroma, pseudomalignant osseous tumors, synovial chondromatosis, and synovial sarcoma should be reserved for discrete soft tissue mass showing calcification and ossification.[31] Myositis ossificans demonstrates a 'zonal phenomenon' of peripheral calcification. It is often inhomogeneous or amorphous in its early stages and may increase or decrease in size in few weeks. [24,29,30] Extraskeletal chondroma and enchondral ossification shows chondrogenic or chondroblastic differentiation of the inner part of mass, with a distinct cartilage cap and fibrous capsule.[13] Synovial osteochondromatoses usually have multiple osteochondrous nodules of synovium with loose bodies in the joint. [4,13,32] Tumor calcinosis is a well-defined calcified mass which shows layering when imaged with a horizontal beam. [4,13,33] Synovial sarcoma may show calcification but may have a coexistent adjacent bony lesion, and is cytologically active and atypical. [16,34] Chondrosarcoma and osteosarcoma have scattered, patchy, amorphous calcification with distinct clinicalhistological findings.[4, 13]

However, in the present case, the lesion originated at the infrapatellar or intercondylar fat pad, growing through the anterior aspect of the joint, as described by Milgram and Dunn, [9] Dienst et al. [24] and Oliva et al. [5] The present case is unique as no such large para-articular osteochondroma extending on both sides of the patellar tendon simulating femoral condyle with a groove has been described in the literature. We are of the opinion that the groove might have been formed due to the constant gliding of the patellar tendon over the osteochondroma over the previous 15 years. The patient did not experience extensive restriction of knee flexion due to the stretch elongation of the quadriceps mechanism and the capsule.

In conclusion, we hypothesize that the tumor remained asymptomatic for such a long period because of its slow growth, stretch elongation of the quadriceps mechanism and capsule.

Conflicts of Interest: No conflicts declared.

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