Rare retro-patellar unilateral multiple osteochondroma in patellar tendon: a case report

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#### 1. INTRODUCTION

Osteochondromas (OC) are benign tumors that account for 20-50% of benign and 10% of all bone tumors. The incidence rate of OC in males is twice that of females and its peak is usually in the second decade of life (1). These lesions are inherited in an autosomal dominant manner and cause the formation of isolated lesions or multiple exostoses during the development of bones in the process of enchondral ossification in the long bones (2). Also, OC is associated with the mutation of some tumor suppressor genes, including EXT1 or EXT2 genes. OC have a periosteal origin and are formed in the active parts of bones, including the metaphysis of long bones and the cartilage at their ends. Studies show that OC can be caused after surgery/radiation-induced injury and hematopoietic stem cell transplantation (3).

Osteocartilaginous exostosis is mainly detected in childhood and in the form of palpable masses, with chronic pain and sometimes with edema (4). The presence of misplaced bone masses in the joint capsule and sometimes with cartilaginous coating in radiographic images is one of its common manifestations. The most common joints involved in OC are the hip and knee joints. Patellar OC is rare and mostly affects the patellar bursa. Moraes et al. (2014) reported a patellar OC measuring  $8 \times 6 \times 3$  cm anterior to the patella in a 60-year-old man who was painless and without limitation of flexion-extension in the knee joint (5). In the present case, rare retro-patellar OC was observed in the area of the patellar ligament.

## 2. CASE REPORT

A 19-year-old male presented with swelling, chronic pain and a palpable mass in the anteroinferior region of the left knee, and after examination with the help of lateral radiography of the left knee (Figure 1), with irregularities in the inferior of patella joint surface and multiple exostosis was observed in the patellar tendon region. The tumor lesion had sharp borders and a hyper-dense appearance, and no signs of fracture or dislocation were observed in the tibiofemoral and patellofemoral joints.

After surgery of the left knee joint, radical resection of the mass and proximal osteotomy of the left tibia, 3 irregular bone masses with total dimensions of  $7 \times 5 \times 1$  cm were sent for histopathological examination. In histopathological evaluation, well-differentiated trabeculae-spongy bone tissue, containing proliferated chondroid and osteoid areas in hypodense lacunae which covered with fibro-fatty tissue and thick hyaline cartilage was observed (Figure 2). This evaluation also confirmed multiple exostosis in the patellar tendon. After 3 months of follow-up, the patient had no movement disorders related to flexion and extension of the knee joint, pain and tumor lesion.

# 3. DISCUSSION

OC is a benign tumor of bone tissue that occurs mainly during endochondral ossification of growth plates

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of long bones. OC is often caused by hereditary multiple exostoses in the areas of the tibiofemoral and patellofemoral joints (6). Of course, this lesion is rarely observed in the patellar ligament and usually causes stiffness of the knee joint (joint locking), dysfunction of the anatomic function of the quadriceps femoris muscle tendon and loss of knee joint extension, edema, chronic pain, ossification of the ligament, formation of fibro- fatty tissue, degenerative arthritis and finally neurovascular compression (7).

In the present case, the patient presented with common symptoms of OC, including edema, chronic pain and stiffness in the knee joint and palpation of solid masses in the patellar ligament. These symptoms could be due to the inflammatory processes involved in the degeneration of dense connective tissue around the mass of exostoses in the lacunae of the patellar ligament. Studies show that OC is associated with good prognosis and low risk of metastasis, but due to the disturbance in the anatomical alignment of the knee joint, the masses are removed by radical resection and the patellar ligament is fixed and stabilized at the connection to the tibial tuberosity (5). In the report of Pandian et al. (2016), in a 22-year-old man, bilateral OC masses in the patella and patellar ligament caused severe arthritis, chronic pain, edema, and impaired flexion-extension knee movements (8). In 15% of cases, the mass is surrounded by a calcified cartilage cap around the bony lacuna, and its thickness even reaches >15 mm (4). Studies show that the incidence ranges of OC are 0.9 - 1.4% per 100,000, which is rare in the patellar ligament. If there is no intervention, valgus deformity in the ankle, knee and pelvic joints is expected in the third decade of life. However, vascular compression and/or nerve compression along with acute pain, extensive edema and tissue degeneration are observed in OC of the hip (sacroiliac-iliofemoral) and shoulder (glenohumerus) joints (9 and 10).

#### 4. CONCLUSION

OC is a benign tumor of the articular-metaphysic areas of long bones that rarely causes intra-articular and intra-ligamentous lesions. OC patellofemoral joint is rare and mainly causes limitation of flexion/extension movements of the knee joint along with stiffness and chronic pain in the area, which can be treated with radical resection of the mass.

## **AUTHOR CONTRIBUTIONS**

Mohsen Akbaribazm: Conceptualization; writing – original draft; writing – review and editing. Hossein Pirmohamadi: Conceptualization, supervision, writing – review and editing. Mohsen Rahimi and Mohammad Kazem Emami Meibodi: Supervision, writing – review and editing.

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# CONFLICT OF INTEREST STATEMENT

The authors declare that there is no conflict of interest.

### CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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# Figure legends:

**Figure 1:** X-ray image of the left knee joint. F: Femur, P: Patella, PL: Patellar ligament, Fi: Fibula, T: Tibia, TT: Tibial tuberosity and OC: Osteochondromas masses.

Figure 2: Histopathology of osteochondromas masses. FFT: Fibro- fatty tissue, T: Bone trabeculae (bone matrix-calcified region), L: Osteocytic lacunae. (H&E staining  $\times$  100, Scale bar = 200  $\mu$ m).



