Case Report

Unusual presentation of osteochondroma (Trevor's disease)

Digvijay Agarwal, Rajesh Maheshwari, Atul Agrawal

Department of Orthopaedics, Himalayan Institute of Medical Sciences, Jollygrant, Dehradun, Uttarakhand, India

Address for correspondence:
Dr. Digvijay Agarwal,
Department of Orthopaedics, Swami Ram Himalayan University, Jollygrant, Dehradun, Uttarakhand, India.
E-mail: digidigi5@yahoo.co.in

ABSTRACT

Osteochondromas are common benign tumors. They probably are developmental malformation rather than true neoplasm and are thought to originate within the periosteum and small cartilage nodule. They rarely develop in the joint. Trevor disease or dysplasia epiphysealis hemimelica (DEH) refers to intraarticular epiphyseal osteochondroma. DEH has an incidence of 1 in 1 million and is characterized by asymmetric overgrowth of cartilage. Though many cases of DEH are asymptomatic, but they may be troublesome when they cause mechanical and pressure symptoms depending on the size and location. We report a case of Trevor's disease of the knee in a 21-year-old male with mechanical obstruction.

Keywords: *Asymptomatic, osteochondroma, Trevor's disease*

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Introduction

Osteochondromas are common benign tumors. They probably are developmental malformation rather than true neoplasm and are thought to originate within the periosteum and small cartilage nodule. Osteochondroma may occur on any bone preformed in cartilage but usually are focused on the metaphysis of the long bones near the physis. They are seen most often on the distal femur, proximal tibia, and proximal humerus. They rarely develop in the joint. Trevor disease or dysplasia epiphysealis hemimelica (DEH) refers to intraarticular epiphyseal osteochondroma.[1] Although first reported by Mouchet and Belot in 1926 and called "tarsomégalie,"[2] it was delineated as a distinct entity by Trevor in 1950.[3] In 1956, Fairbank[4] described the characteristic involvement of either the lateral or medial half of a single limb. He first used the term "DEH," which appears to be the most logical and frequently used nomenclature. DEH is a rare disease characterized by asymmetric overgrowth of cartilage. Though many cases of DEH are asymptomatic, but they may be troublesome when they cause mechanical and pressure symptoms depending upon the size and location.[1]

We report a rare and atypical presentation of Trevor's disease affecting knee in a 21-year-old male.

Case Report

A 21-year-old male came to our outpatient department with a chief complaint of a restricted range of motion right knee since one and a half years. The patient gave a history of difficulty in bending right knee, which gradually increased with time. However, there is no history of any injury or overuse symptoms. Physical examination revealed no gait disturbance. Bony prominences were palpable over the medial femoral condyle and proximal tibia. No tenderness was elicited around the knee. The range of motion was restricted with only 80° of flexion with no extension lag [Figure 1a]. Knee was found to be stable with an exception of positive McMurrays test for the medial meniscus. Radiological evaluation with X-rays [Figure 2] were suggestive of multiple exostosis of right femur, tibia, fibula, and ribs with a radiodense structure in

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the intercondylar region, whereas computed tomography scan [Figure 3a and b] was suggestive of bony outgrowth in continuation with the cortex of medial condyle seen in the intercondylar fossa of right femur measuring approximately 3.3 cm, no cortical breach was seen. The patient was planned for surgical excision and biopsy of the growth. The patient was approached via midline posterior incision. On exploration, swelling was found to be originating from the lateral surface of the medial femoral condyle, protruding in the intraarticular region, closely encasing the posterior cruciate ligament. On exploring the growth after detaching it was found that there was a cartilaginous covering present over the resected mass. Microscopic examination of the biopsy tissue suggested tissue was capped with cartilage. Beneath, the cartilage mature bony trabeculae rimmed by osteoblasts was seen separated by intratrabecular tissue with no dysplastic changes; thus, the suggestive of osteochondroma [Figure 4]. Postoperatively, the patient improved symptomatically with a full range of motion [Figure 1b] with no mechanical obstruction.

Figure 1: (a) Preoperatively range of motion right knee 0–80. (b) Postoperatively almost full range of motion

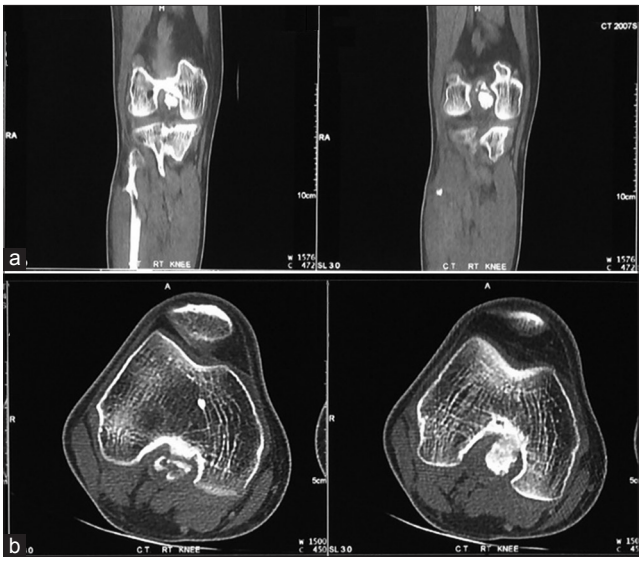

Figure 3: (a) Coronal section of computed tomography scan of the right knee showing bony outgrowth in the intercondylar fossa of right femur. (b) Computed tomography scan transverse section right knee showing bony outgrowth in continuation with the cortex in the intercondylar fossa of right femur

Discussion

Trevor's disease is commonly seen in males, with age group from 2 to 14 years, common location in knee and ankle. The disease is very rare and has an incidence of 1 in 1 million.[5] However, this estimate may be artificially low since many cases may go unrecognized because of a similar histologic appearance to osteochondroma or because the lesion may be asymptomatic.[6,7] In our case, the patient presented at the age of 21 years which does not coincide with the probable age group for Trevor's disease. This may be because the disease usually remains asymptomatic for a long time and may become troublesome when they cause mechanical and pressure symptoms.[1] In our case, the patient presented with mechanical obstruction of the knee joint. Initially, the case mimicked as a case of internal derangement of the knee or intraarticular loose body, which may

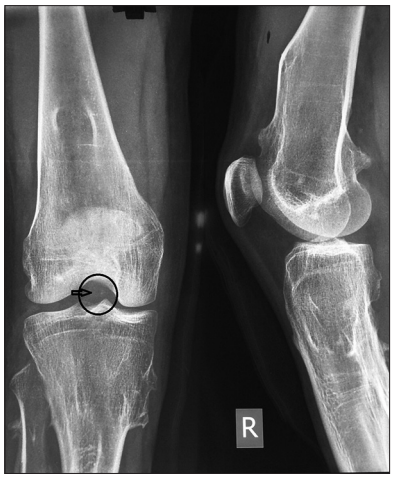

Figure 2: X-ray of right knee anterior-posterior and lateral view. In anterior-posterior view, arrow shows a radiodense structure in the intercondylar region

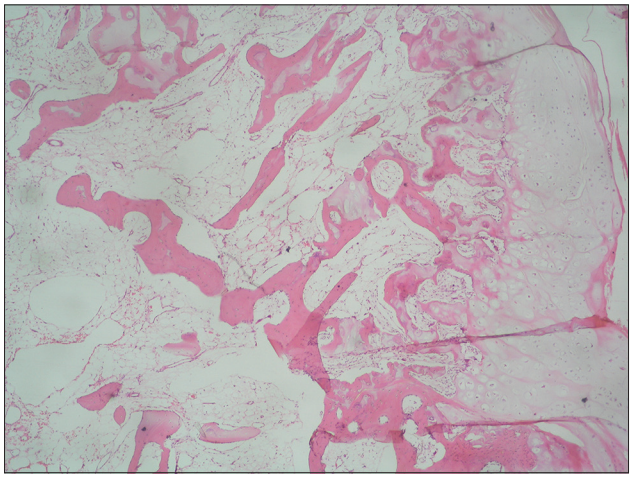

Figure 4: Histopathology of resected mass showing tissue capped with cartilage with bony trabaeculae rimmed by osteoblast, separated by intertrabecular tissue with no dysplatic changes. Suggestive of osteochondroma

be because of the intraarticular location of the growth. Such cases should be evaluated properly, so as to differentiate it from synovial chondromatosis, osteochondroma, large osteophyte and pigmented villonodular synovitis, and meniscal pathology. Diagnosis of the case was based on the proper history, physical examination, radiological investigations, and histopathology. Because of similar pathoanatomy and histologic features of DEH and osteochondroma, the decision to classify lesion as DEH is based primarily on the intraarticular location of the lesion.[4] In literature, the treatment ranges from simple observation of asymptomatic lesions to surgical excision. Good results have been seen with nonoperative treatment in the literature. The treatment should be individualized depending upon the clinical findings. Asymptomatic lesions can be treated conservatively as no malignant transformation is seen. Surgical intervention should be done if lesion produces pain, deformity, or any joint obstruction. Literature suggests that excellent results have been seen with an excision of juxta-articular lesions, but only fair or poor results with the intraarticular lesion. Excision of intraarticular lesion has a high risk of osteoarthritis.[8] We performed surgical excision in our case and patient was completely relieved of his symptoms with an almost full range of motion postoperatively. But such cases should be closely observed in follow-up to look for any osteoarthritic changes, which may arise due to cartilage damage/joint incongruity.

Conclusion

Osteochondromas are commonly found in the metaphyseal region of the long bones. However, intraarticular presentation (Trevor's disease) should be considered in the patients who present with the symptoms of mechanical obstruction or internal derangement of the knee. These benign tumors are a very rare possibility, but still these tumors can develop at this very unusual site. Because of asymptomatic nature of the disease, the patient can present even in adult age group, so the differential diagnosis of Trevor's disease should always be considered in adult age group. Surgical excision is the treatment of choice in all symptomatic patients.

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Conflicts of interest
There are no conflicts of interest.

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