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MALIGNANT TRANSFORMATION OF A SOLITARY OSTEOCHONDROMA OF DISTAL TIBIA INTO SECONDARY CHONDROSARCOMA: A CASE REPORT

Orthopaedics

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ABSTRACT

Osteochondroma is most common benign bone lesion commonly arising from the metaphysis of long bones and covered by a cartilaginous cap. Usually osteochondromas are asymptomatic, painless, and discovered incidentally. The common causes of pain in osteochondroma include bursal inflammation, stalk fracture, compression of neurovascular systems, malignant transformation. Although malignant transition to chondrosarcoma is uncommon (less than 1% in solitary cases and upto10% in hereditary multiple exostoses), patients who come after skeletal maturity may be investigated if they experience abrupt increases in size and increased discomfort. We present a case of 56-year-old male presented with history of low energy trauma and complaints of progressive swelling over distal 1/3rd of right leg for 25 years. Examination revealed bony hard swelling having ill-defined margins. Radiological examination showed an ill-defined, sessile lesion arising from the lateral cortex of distal tibia and causing pressure erosion of distal fibula. Patient was operated for wide resection of the mass and intramedullary interlocking nailing for tibia fracture. Grossly, mass was sessile with cauliflower-like appearance. Histopathology confirmed a low-grade secondary chondrosarcoma, managed with post operative radiotherapy. Patient was followed up for 6 months postoperatively and has shown functional and radiological improvement with no evidence of recurrence.

KEYWORDS

Osteochondroma, Ankle, Cartilage Cap, Secondary Chondrosarcoma, Radiotherapy.

INTRODUCTION:

Osteochondroma is the most common benign tumour, making about 20% to 50% of all benign tumours. It is characterized by cartilage-capped bony protrusion, seen commonly during childhood and adolescence. (1)

Usually osteochondromas are asymptomatic, painless, and discovered incidentally. The most common causes of pain in osteochondroma include bursal inflammation, stalk fracture, and compression of nearby neurovascular systems, malignant transformation. Malignant transformation is very rare and found in less than 1% of reported cases in solitary osteochondroma and approximately 10% in hereditary multiple exostoses. (2)

Proximal humerus, scapula, proximal femur and pelvis are the common locations where transformations are reported. (3) Signs of a potential malignant change include pain, progressive growth after skeletal maturity, ill-defined tumour margin, internal lucency, permeation into neighbouring bone, and a cartilage cap thickness of more than 2 cm. (4,5)

Osteochondromas arising from the interosseous border of the distal tibia and involving distal fibula are very rare. Considering its proximity to the ankle joint, early excision of this deforming distal tibial osteochondroma is done to avoid the future risk of ankle deformities and syndesmotic complications.

Here we present a rare case of malignant transformation of osteochondroma of distal tibia with associated fracture in an adult with most of the clinical and radiological signs of transformation, in whom histopathology confirmed a low-grade secondary chondrosarcoma, managed with wide excision of tumour and osteosynthesis along with post operative radiotherapy.

Case:

56-year-old male presented with alleged history of low energy trauma and complaints of pain, swelling and deformity in right leg. History of progressive swelling over distal 1/3rd of right leg for 25 years, insidious in onset and painful on exertion only. Examination revealed bony hard swelling of size 12 x 6 cm, overlying distal tibia having ill-defined margins, irregular thickened surface and fixity to bone. No distal neurovascular deficit noted. (Figure 1)

Plain Radiograph and CT scan of right leg revealed an ill-defined, sessile cartilaginous exostotic lesion with lucent areas in between, arising from the lateral cortex of distal tibia and causing pressure erosion of distal fibula along with distal 1/3rd of tibia fibula fracture. (Figure 2&3)

Patient was posted for wide resection of the mass and intramedullary interlocking nailing for tibia fracture. Grossly, the mass was of size 8x3x1cm, lobulated, sessile with cauliflower-like appearance and consist of multiple shiny grey white firm to hard piece of tissue with glistening capsule having thickness of >2 cms. (Figure 4)

Microscopic examination showed abundant cartilaginous matrix with chondrocytes embedded in lacunae showing moderate cellularity separated by myxoid stroma; cells showed mild nuclear atypia with areas of calcification, suggestive of Chondrosarcoma of low grade.

Post Operatively, PET scan was performed and in view of residual ill-defined soft tissue lesion with permeative lytic pattern of size approx. 25x33x77mm, radiotherapy was given for period of 2 months in dose of 50 Gy in 25 fractions at 200cGy per fraction.

At 6-month follow-up, the patient is asymptomatic, with full range of ankle movements, and no clinical or radiological signs of recurrence. (Figure 5 & 6)

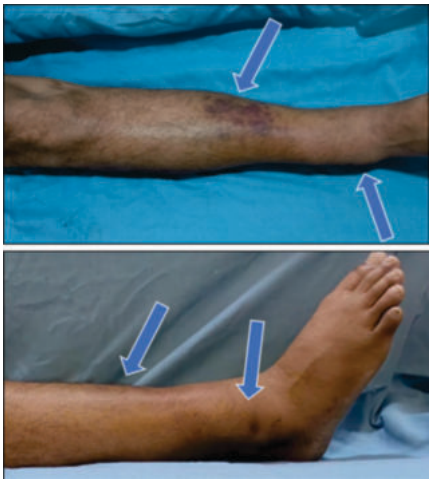


Figure 1- Swelling over distal leg (Right)

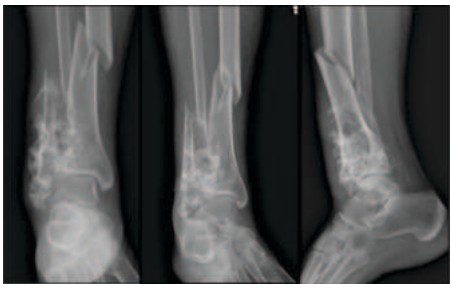


Figure 2 – Fracture with sessile mass involving distal tibia fibula (Xray Right Ankle)

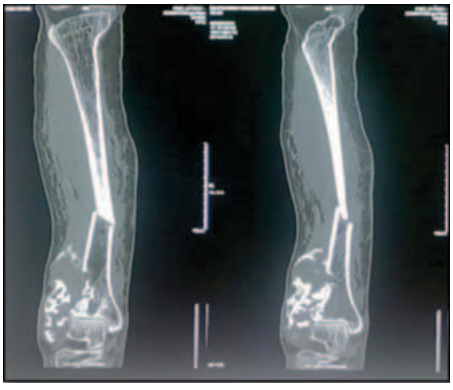


Figure 3 – Fracture with sessile mass involving distal tibia fibula (CT scan Right leg)

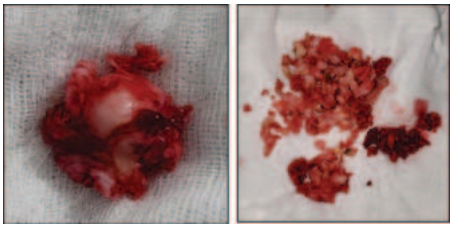
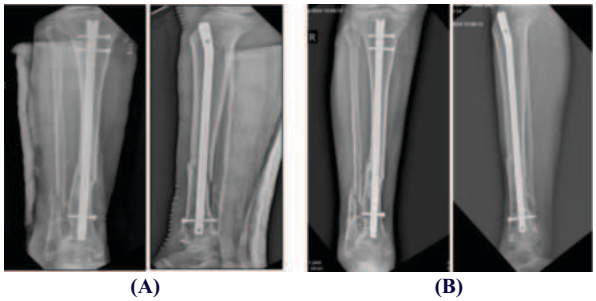


Figure 4 – Gross appearance of excised tumour with cartilage cap (thickness >2 cms)



(A) **(B)**

Figure 5 – Immediate post op X rays(A) and 6 months follow up X rays(B) in A/P and lateral views.

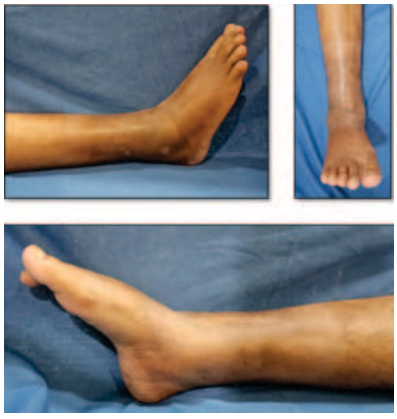


Figure 6 – Clinical condition and range of motion, 6 months post operatively.

DISCUSSION:

Osteochondroma is most common benign bone lesion commonly arising from the metaphysis of long bones and composed of spongy bone covered by a cartilaginous cap. (6)

The metaphysis of proximal tibia, distal femur, proximal femur and proximal hue the most commonly affected sites, though they can occur at all bony sites. (7,8)

Osteochondromas may be asymptomatic or present as a mass or bony lump, when they are discovered incidentally. Progressive expansion can occasionally result in fractures and pressure symptoms including skeletal deformities or nerve compression. Although malignant transition to chondrosarcoma is uncommon (less than 1% in solitary cases and more in multiple), patients who come after skeletal maturity may be investigated if they experience abrupt increases in lesion size and increased discomfort. (9)

Although they have been documented in the literature, osteochondromas originating from the distal tibia's interosseous border are uncommon. If left untreated, these tumours can have a "mass effect" that includes degenerative alterations in the ankle, syndesmotic issues (diastasis or synostosis), varus/valgus deformities of the ankle, mechanical obstruction of joint motion, and plastic deformation of the lower end of the tibia and fibula. (7,8) So most of the authors favour surgical resection as preferred treatment for osteochondromas located in this area. (10,11) There is still little information of the natural evolution after treatment of osteochondromas arising from the distal aspect of either the tibia or the fibula. (10)

Resection is recommended for patients whose lesion is causing a cosmetic deformity or possible harm to nearby joints or neurovascular structures, for a lesion that has characteristics of malignant transformation, for a lesion in an area that has experienced minor trauma, or for a lesion that is causing symptoms due to irritation of the surrounding soft tissue. (12)

In order to avoid recurrence, Mirra emphasized the significance of total cartilaginous cap excision. (13) The literature has described anterior, posterior, and trans-fibular approaches. According to Wani et al., the anterior approach in this instance is linked to the lowest level of postoperative morbidity. (14)

A rare type of tumour known as secondary chondrosarcoma arises when an existing cartilaginous lesion undergoes malignant transformation. A malignant change into secondary chondrosarcoma may be indicated by a fast expansion of the osteochondroma's cartilaginous cap. Most occurrences of secondary chondrosarcoma fall into the low to intermediate grade category. Most patients have a good prognosis, and distant metastases are uncommon. The five-year overall survival rate is approximately 90%. Although large margin surgical resection is thought to be the best course of treatment, 10% to 20% of patients still experience local recurrence, which is a significant issue. (15)

CONCLUSION:

It is very rare for a solitary osteochondroma to turn malignant, most common malignancy is secondary chondrosarcoma, however mostly it remains low grade with favourable prognosis. Although it is very difficult to diagnose the transformation, in the event of suspicious clinical and imaging features, a wide excision and histopathological confirmation are mandatory. Marginal resection leads to a high recurrence, usually appearing within 5 years.

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