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Challenges and Solutions in Managing Recurrent Distal Tibia Interosseous Osteochondroma: A Case Study and Review of Literature

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Learning Point of the Article:

Osteochondroma can present in unusual locations such as the distal tibia with fibular deformation and ankle varus deformity. Observation is suitable for asymptomatic lesions, while symptomatic cases or those with concerning imaging findings may require surgery. Complete resection is important to minimize relapse and risk of malignant transformation. Careful consideration is warranted in pediatric cases to prevent growth plate damage.

Abstract

Introduction: Osteochondroma, a common benign bone tumor, predominantly affects young individuals, with a higher prevalence in males. It typically manifests as a bony growth capped with cartilage near bone growth plates, often extending away from joints. While most cases are asymptomatic, some may present with pain, swelling, or mechanical complications necessitating surgical intervention. Recent research implicates genetic mutations, particularly in the EXT-1 gene, in osteochondroma development, with homozygous EXT1 deletion commonly found in sporadic cases.

Case Report: A 15-year-old girl presented to out patient department with recurrent osteochondroma in the distal tibia, an unusual location, with complications such as fibular deformation and ankle varus deformity. Initial surgery was performed elsewhere through an anterolateral approach, but the patient experienced persistent pain and serous discharge from the surgical scar. Examination revealed an unhealed scar with a discharging sinus and a firm, bony swelling on the left lower leg, along with a reduced range of motion. X-ray and MRI confirmed a pedunculated mass emerging from the distal tibial metaphysis, causing extraneous compression and deformity of the fibula without intraosseous infiltration. Histopathological examination of the resected specimen confirmed osteochondroma recurrence. Surgical management involved en bloc resection of the tumor and perichondrium through a posterolateral approach, with subsequent debridement of the previous surgical wound. Post-operative recovery was uneventful. Literature review indicates that observation is suitable for asymptomatic lesions, while symptomatic cases or those with concerning imaging findings may require surgery. Complications following surgical excision are reported, with recurrence rates ranging from 2% to 11.6%, highlighting the importance of complete resection to minimize relapse and risk of malignant transformation. Careful consideration is warranted in pediatric cases to prevent growth plate damage.

Conclusion: Osteochondroma management involves tailored surgical intervention based on symptoms and imaging findings, with complete resection recommended to optimize outcomes and minimize recurrence, particularly in pediatric patients.

Keywords: Osteochondroma, benign bone tumor, distal tibia, recurrent osteochondroma, interosseous osteochondroma.

Introduction

Osteochondroma is a common benign bone tumor, accounting for a significant portion of benign bone tumors and a smaller fraction of all bone tumors. It typically occurs in young individuals, with a higher prevalence among males, and is characterized by a bony growth capped with cartilage on the

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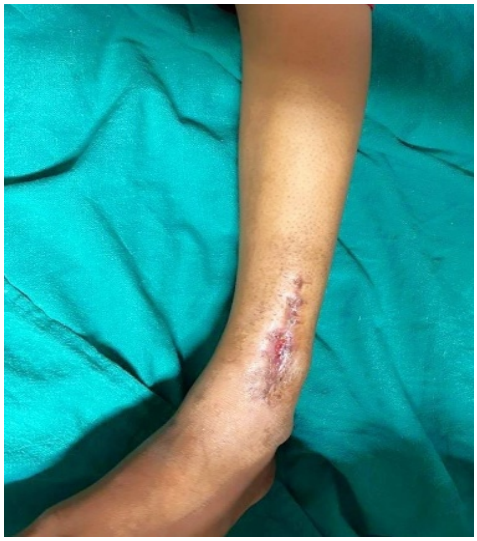

Figure 1: Clinical image showing previous surgical scar mark with a discharging sinus over the anterolateral aspect of left lower leg.

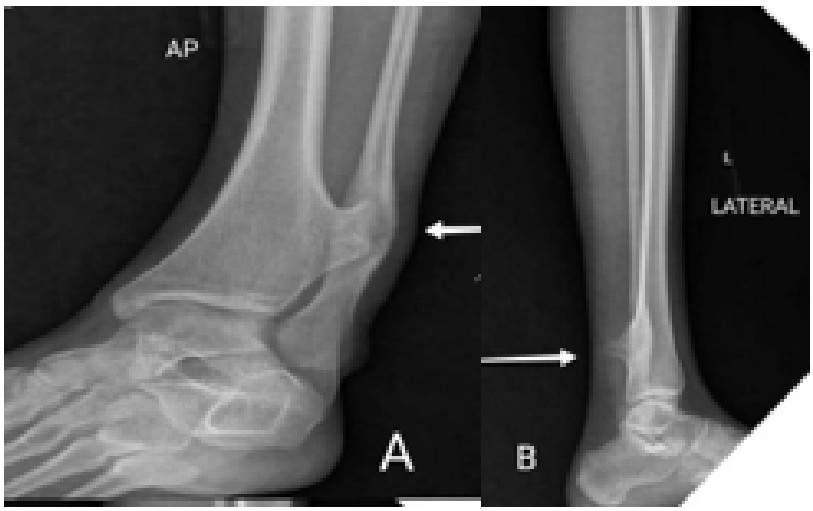

Figure 2: Pre-operative plain radiograph ankle (A) anteroposterior and (B) lateral view, showing pedunculated mass emerging from the metaphysis of the distal tibia.

outer surface of a bone, usually near the growth plate and extending away from the joint [1-4]. About half of these growths develop around the knee joint [5], primarily affecting long bones such as the femur, tibia, and humerus, with the distal femur being the most involved site [6]. While most osteochondromas do not cause symptoms and are often discovered incidentally [2, 4], some may lead to issues such as mechanical compression of nearby structures, fractures, or deformities, necessitating surgical removal if symptomatic or if there are suspicious findings on imaging [7].

Recent research has shown that genetic mutations, particularly in the EXT-1 gene, play a role in the development of osteochondromas [1, 8]. Homozygous deletion of EXT1 is commonly found in sporadic cases of these tumors [9, 10].

Management typically involves observation for asymptomatic lesions, while symptomatic ones or those with concerning imaging features may require surgical intervention. Recurrence following surgical removal is rare, occurring in only a small percentage of cases [11].

A case is presented here involving a 15-year-old girl with a recurrent osteochondroma located unusually in the distal tibia, leading to complications such as deformation of the fibula, ankle varus deformity, and involvement of the distal interosseous membrane due to mechanical reasons.

Case Report

A 15-year-old girl visited the outpatient department of a tertiary

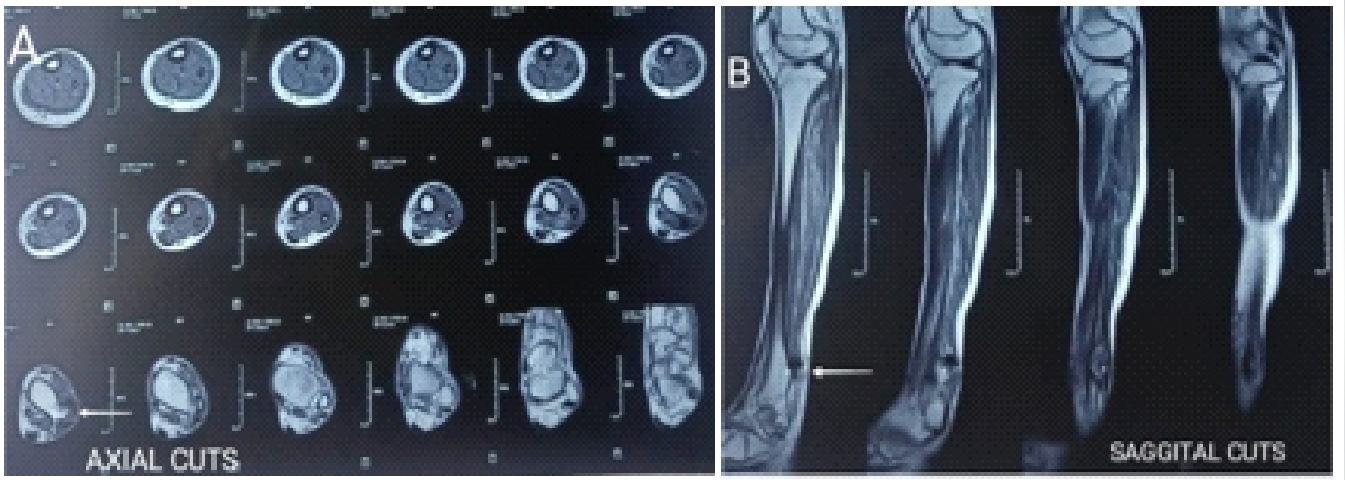

Figure 3: MRI leg with ankle joint (A) Axial cuts (B) Sagittal cuts – showing solitary osteochondroma of the outer margin of lower tibial metaphysis as broad stalk exostosis with continuity of bone cortex and medullary marrow space to the host bone.

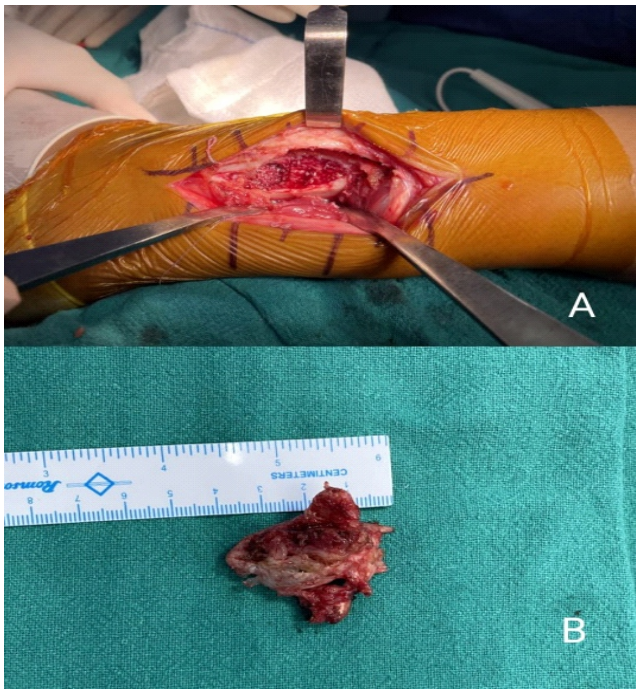

Figure 4: Intraoperative image (A) following en bloc tumor resection of tumor, also showing deformed fibula and (B) the resected lesion 3x2x1 cm in size, grey-brown to grey-white tissue.

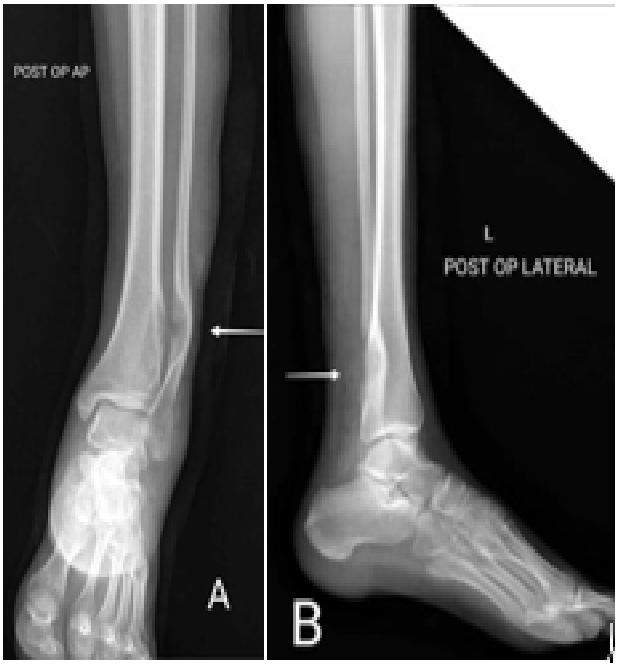

Figure 5: Post-operative plain radiograph ankle with leg (A) anteroposterior and (B) lateral view.

care hospital, complaining of recurring pain and swelling in her left lower leg for the past 4 months. The pain worsened when her leg was moved passively. She had undergone surgery for the same issue 4 months ago at a private hospital, where an excisional biopsy was performed through an anterolateral approach. In addition, she reported intermittent serous discharge from the surgical scar since then, with no history of significant trauma, past illnesses, or medication use (Fig. 1).

On examination, an unhealed scar with a discharging sinus was observed on the anterolateral aspect of her left lower leg. There was also a poorly defined, round swelling measuring approximately 3 × 2 cm on the posterolateral aspect of the same leg. No discoloration or dilated veins were noted. On palpation, the swelling felt firm and bony with indistinct edges. It was immobile and non-tender. Local temperature and pulsations were normal, and there were no signs of increased temperature or pulsation. The patient had full range of motion in her knee, ankle, and toes, and pulses in the dorsalis pedis and posterior tibial arteries were intact. Neurological examination of the limb revealed no abnormalities.

Laboratory findings showed normal hemoglobin levels (12 gm%), with erythrocyte sedimentation rate (ESR) at 10 and C-reactive protein (CRP) at 0.18. Gram stain and culture sensitivity of the serous discharge yielded no growth.

X-ray examination revealed (Fig. 2) a distinct, pedunculated mass emerging from the metaphysis of the distal tibia, extending away from the joint and abutting the distal fibula. The lesion measured 3 × 2 cm and displayed features consistent with both cortical and medullary components, indicating continuity with the underlying bone. In addition, the examination revealed distortion of the distal fibula and deviation of the ankle joint towards the varus, with the degree of deviation measured at 20°.

MRI revealed (Fig. 3) solitary osteochondroma of the outer margin of lower tibial metaphysis as broad stalk exostosis showing continuity of bone cortex and medullary marrow space to the host bone. This exostosis of 29 mm length and 19 × 10 mm transverse span caused extraneous chronic pressure compression and deformity of the lower fibula without any intraosseous infiltration. The tip of the lesion was surrounded by a 1 mm thin cartilaginous cap. No imaging feature for malignant transformation was reported. Extraneous compression/stretching of the peroneal vessel along with stretching of flexor hallucis longus and peroneus muscle was seen. Sheet-like soft-tissue edema of the overlying superficial subcutaneous plane was also reported.

Results of metabolic and radiological workup could finally ascertain that there is a recurrence in a previously operated case of osteochondroma of the distal tibia. The patient underwent en bloc resection of the tumor along with removal of the cartilage cap and perichondrium through posterolateral approach (Fig. 4) and debridement of the previous surgical wound present anterolaterally. The resected specimen was sent for histopathological examination. The post-operative period

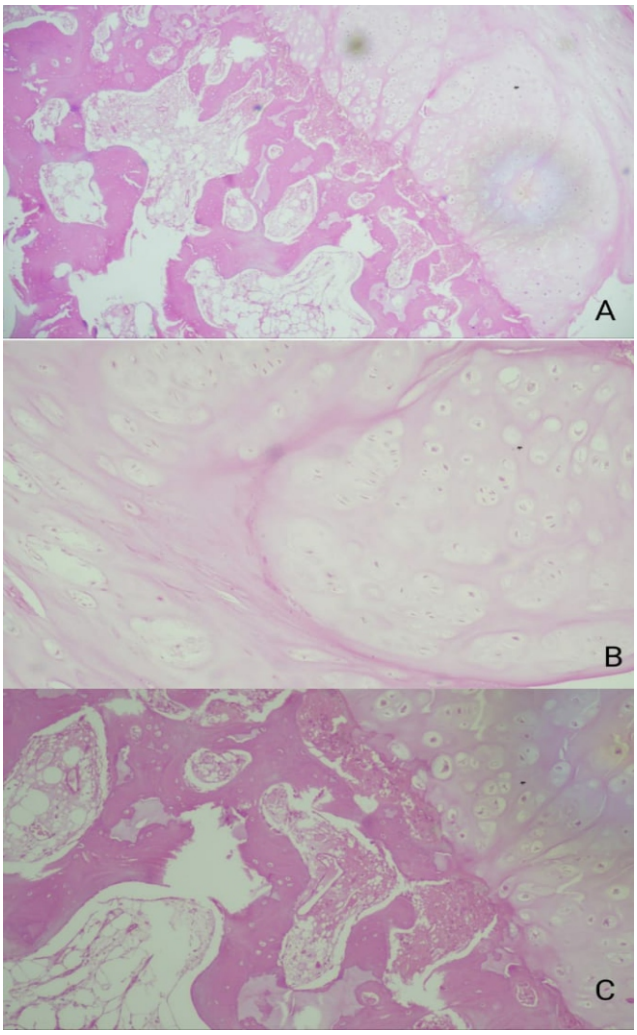

Figure 6: Histopathologic examination. (A) Microscopic pictograph shows cartilaginous tumor, with a cartilaginous cap with an underlying bony cavity. (H & E, 200×). (B) Outer fibrous perichondrium with underlying cartilage cap showing the distribution of chondrocytes, surrounded by cartilaginous hyaline matrix. (H & E, 400×). (C) The overlying perichondrium continuing with the periosteum of the underlying bone. (H & E, 400×).

was uneventful (Fig. 5). After the operation, a non-weight bearing below knee slab was applied. The patient was followed up in OPD with physical examination and radiography. At 6 month follow-up, she has completely recovered and there is the return of full ankle functions.

Histopathology (Fig. 6) revealed on gross examination grey-brown to grey-white tissue with no areas of necrosis and hemorrhage. The specimen showed a benign cartilaginous tumor comprising of the cartilaginous cap with an underlying bone marrow cavity continuing with the marrow cavity of the adjacent bone and showing scant cellular marrow elements and fibrosis. The tumor comprised three layers, an outer fibrous perichondrium with an underlying cartilage cap showing a distribution of chondrocytes, surrounded by a cartilaginous hyaline matrix. The overlying perichondrium is seen continuing with the periosteum of the underlying bone. There was no evidence of any fibrillary cartilage matrix, loss of architecture, nodularity, myxoid changes, or any wide fibrous bands identified in the sections examined. The features were thus consistent with osteochondroma confirming the diagnosis of recurrence.

Discussion

Cortical and marrow continuity between the lesion and the parent bone, and a cartilage cap are the key radiological features of osteochondroma [12]. If osteochondroma is located at the metaphysis of long bones, it is usually diagnosed only by radiographs [1]. MRI/MDCT can be used to delineate complex lesions or lesions situated at complex anatomical regions like the shoulder girdle, pelvis, or spine [13]. Tumor growth after skeletal maturity, irregular or lobulated margins, irregular or scattered calcifications, internal lytic areas, and erosion or destruction of adjacent bones are some of the worrisome imaging features [14-17]. MRI is the best method for imaging the exact morphology of a tumor and all the reported complications such as compression of the spinal cord and nerves, bursitis and muscular impingement, and vascular complications [4, 18-21]. A sign of malignant transformation is a cartilage cap thickness greater than 3 cm in children or 2 cm in adults [7, 20, 22, 23].

Observation initially with plain radiographs and subsequently by clinical examination is generally, the mainstay of conservative treatment [24]. If pain develops or the tumor enlarges after the closure of growth plates, an MRI may be needed for further investigation [25]. Surgical management is indicated in patients with severe pain, mechanical compression of adjacent structures, fracture, osseous deformities, bursitis, vascular or neurological complications such as tingling, numbness or loss of pulses, cosmetic reasons, increased risk of malignant transformation, and uncertain diagnosis [4, 12, 24]. In a few cases, osteochondromas can be located under tendons, creating pain during relevant movements. Finally, the stalk of a pedunculated osteochondroma can snap due to an injury causing pain and swelling. Surgery should not be carried out in cases of asymptomatic tumors, as the risk of surgery-related complications is higher than tumor-related ones [4].

In a study done by Wirganowicz et al., they described the surgical risk for the elective excision of 285 osteochondroma; they found the complication rate to be 12.5%, among which the most common was neurapraxia, followed by arterial laceration, compartment syndrome, and fibula fracture [26].

Bottner et al. reported a recurrence rate of 11.6% after evaluating the surgical treatment of symptomatic osteochondromas in 86 patients. Major and minor

complications occurred in 4.7% and 7% of patients, respectively [27].

Florez et al. studied 113 solitary osteochondromas treated between 1970 and 2002. In 2 patients tumors transformed to secondary chondrosarcomas while in six patients the tumors recurred. The authors noted that the recurrence of exostoses is rare and estimated to be 2% of the resections [11]. Complications vary between 11.6% and 12.5% after surgical removal [26,27].

Based on surgeries done for distal tibial interosseous osteochondroma multiple studies are done. For resection of tumors, different methods are used. Different approaches and osteotomy were done and most studies reported a good range of motion and functional outcome postoperatively as compared to pre-operative status. Singh et al. [28] did an anterolateral approach, whereas, Tayara et al. [29] and Pant et al. [30] did a posterolateral approach. An anterior approach was done by Ismail et al. [31] and Wani et al. [32]. All these studies did not do osteotomy. Appy-fedida et al. [33], Gil albarova et al. [34], and Thakur et al. [35] did a trans-fibular approach, fibula osteotomy was included in their surgery. The lateral approach was used by Yang et al. [36], and fibula and Volkman tuberosity osteotomy were done in that study. Complications are reported in both non-osteotomy and osteotomy groups. Although there are fewer studies it is still controversial that in cases of distal tibial interosseous osteochondroma patients, osteotomy should be done or not, and further studies are needed to ascertain the role of osteotomy.

Thus, the recommended intervention is complete resection by removing exostoses at the normal bone base, with consequential removal of the cartilage cap and perichondrium, which we did in our case. The risk of relapse is minimal if the resection bed is clear, and there are no remnants of the cartilage cap and perichondrium, [37]. Apart from that, there is a likelihood of cancerous degeneration in 1% of solitary osteochondromas, therefore, complete resection is obligatory [2, 24, 38].

The relapse rate is probably higher in the immature patient, therefore, in children, resection might preferably be held up until skeletal maturity is reached [39]. To avoid damage to the growth plate, great care must be exercised with tumors close to the physeal plate. Osteochondromas are benign lesions, and thus the prognosis is excellent. Local recurrence is < 2% in the literature [11, 37, 39].

Conclusion

Osteochondromas, though benign, can lead to complications requiring surgical intervention. Complete resection, including removal of the cartilage cap and perichondrium, is recommended to minimize recurrence and risk of malignant transformation. Careful consideration is needed in pediatric cases to avoid growth plate damage. Overall prognosis is excellent with low recurrence rates.

Clinical Message

Distal tibia interosseous osteochondroma can present with complications like fibular deformation and ankle varus deformity. While symptomatic cases or those with worrying imaging results may need surgery, asymptomatic lesions are a good candidate for observation. To reduce the chance of malignant transformation and recurrence, complete resection is crucial.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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