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Telangiectatic osteosarcoma masquerading as aneurysmal bone cyst: A pediatric case complicated by superimposed osteomyelitis

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Abstract

Background: Telangiectatic osteosarcoma (TOS) is an uncommon and aggressive variant of osteosarcoma with numerous blood-filled cystic spaces, leading to confusion with benign lesions like aneurysmal bone cyst (ABC). Early, accurate diagnosis is vital to starting appropriate management and improving overall outcomes.

Case presentation: A 10-year-old boy presented with progressive right knee pain and swelling. Initial imaging revealed an expansile, lytic lesion in the distal femur, suggestive of an aneurysmal bone cyst. Surgical excision and biopsy initially favored telangiectatic osteosarcoma; however, a second histopathology review suggested aneurysmal bone cyst, and cytology showed giant cells and hemorrhage without definitive malignant features. Multidisciplinary review, integrating clinical behavior and imaging findings, confirmed the diagnosis of telangiectatic osteosarcoma. The patient underwent high-dose methotrexate-based chemotherapy. Post-chemotherapy MRI revealed a persistent tumor mass with central intramedullary abscess formation, accompanied by surrounding myositis and sinus tract, consistent with superimposed osteomyelitis. Conservative intravenous antibiotic therapy was initiated successfully. Following infection control, wide local excision of the distal femur with limb reconstruction was performed, achieving oncologic clearance.

Discussion: In summary, this case illustrates the diagnostic dilemma of telangiectatic osteosarcoma, particularly when the histopathologic and imaging findings mimic those of a benign process. Management was further complicated by superimposed infection. Diagnosis can only be achieved through the integration of clinical, radiological, and pathological data.

Conclusion: An oncologically safe resection and limb salvage were achieved with prompt multidisciplinary evaluation and coordinated management. In recognition of the diagnostic pitfalls and possible infectious complications, this approach is relevant to the management of pediatric bone tumors in the setting of cystic imaging findings.

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Introduction

Telangiectatic osteosarcoma (TOS) is a rare and aggressive subtype of osteosarcoma accounting for <4% of all osteosarcomas [1]. It overwhelmingly affects children and adolescents, and on X-ray appears as lytic lesions containing fluid or fluid-fluid levels that can mimic benign bone lesions such as an aneurysmal bone cyst (ABC) [2]. Clinically and microscopically, TOS sometimes resembles ABC, where the diagnosis is challenging and definitive therapy may be postponed. Early distinction between benign and malignant lesions is crucial for appropriate therapeutic planning and improved survival outcomes [3].

Histologically, telangiectatic osteosarcoma is made up of a large number of blood-filled cystic spaces that are lined by tumor cells and are quite different from the bland nature of an actual aneurysmal bone cyst [4]. However, diminutive or inadequately sampled biopsies may show only hemorrhagic spaces and giant cells and cause diagnostic difficulty. These lesions can be distinguished only by immunohistochemical examination and nonspecific results with imaging tests. TOS that requires aggressive multimodal treatment including wide surgical excision and chemotherapy, while ABC is often treated more conservatively, thus misdiagnosis can have dire consequences [5].

Associated with neoplastic bone lesions, osteomyelitis (an infection of the bone) further complicates the clinical scenario. Infections can also mimic clinical and radiologic findings, simulate tumor recurrence, and delay the therapeutic endpoint of cure [6]. In pediatric patients, the presence of a developing skeleton and variations in the immune system make the patients more susceptible to metastatic spread of tumors and infectious complications. Furthermore, surgical treatment such as external fixation for stabilization might serve as a gateway for bacteria into the joint [7].

The case of a telangiectatic osteosarcoma in a child is reported, causing difficulties in diagnosis and management, initially mistaken for an aneurysmal bone cyst and subsequently rendered more difficult by superadded osteomyelitis. It emphasizes the importance of painstaking pathological examination, close evaluation of the imaging, and a vigilant threshold of suspicion of unusual clinical presentation. Moreover, it highlights how infections might affect the natural history of malignancies, which would change the management and, ultimately, the patients' outcomes [8].

Through presentation of this unique clinical case, we sought to enrich the literature on telangiectatic osteosarcoma, emphasize the potential to have a diagnostic pitfall, and contribute to the available literature of the multidisciplinary approach to complex presentations of pediatric oncologic cases. An early detection of concurrent pathologic and infectious processes is important for maximizing therapeutic strategies and increasing the survival rate of affected children [9].

Case presentation

A 10-year-old previously healthy boy presented with progressive pain and swelling of his right knee, which had been gradually worsening for months and remarkably impacted his ambulation. On the initial evaluation right lower limb radiograph revealed a medial radio-opaque lesion involving the distal femur and was suspected for a primary bone tumor as shown in Figure 1. MRI imaging subsequently revealed a well-defined, expansile lesion in the distal meta-diaphysis of the femur, extending up to the epiphyseal region. Radiologically, the lesion had several cystic spaces with fluid-fluid levels, likely consistent with an aneurysmal bone cyst.

Following clinical and radiological assessment, the patient underwent surgical excision of the lesion in addition to the application of an external metallic fixator to stabilize the operated limb. The lesion was initially classified as a telangiectatic variant of osteosarcoma based on histopathological analysis of the excised specimen. Microscopically, showed malignant neoplasm, sparse and dense pleomorphic spindle cells with high nuclear atypia and brisk mitotic activity. Multiple cystic spaces containing blood were noted, separated by thin septa lined by malignant cells. Due to the aggressive nature of histological features, a presumptive diagnosis of telangiectatic osteosarcoma was made.

However, follow-up histopathological review at a tertiary oncology center proposed a diagnosis more in line with an aneurysmal bone cyst. This was characterized as cystic spaces with blood, separated by fibrous septa with bland spindle cells, osteoclast-like giant cells, and hemorrhagic areas, with no definitive evidence of malignant osteoid or sarcomatous material. Cytological analysis was performed on aspirated blood-stained fluid from the lesion for further clarification. Cytology showed scattered giant cells, mononuclear inflammatory cells, histiocytes, and a background of hemorrhage, without invasive malignant cells. We obtained inconclusive results from immunohistochemical studies.

Although cytological and second biopsy results favored a benign process, the aggressive clinical course and imaging appearance raised significant concern for underlying malignancy. After the discussion of a multidisciplinary team correlating all clinical, radiological, and pathological data, the final diagnosis of telangiectatic osteosarcoma was achieved. He was started on systemic chemotherapy according to a high-dose methotrexate regimen for pediatric osteosarcoma. He undergoes chemotherapy with expected toxicities of transient neutropenia, anemia, and mild gastrointestinal disturbance, all controlled with supportive care. Serial serum methotrexate levels were followed and showed appropriate clearance following sodium bicarbonate infusion and leucovorin rescue.

After completing chemotherapy cycles, an external fixator was removed. A follow-up contrast-enhanced MRI of the right femur and knee joint was performed for restaging the disease status, as shown in Figure 2. Imaging studies demonstrated a large, ill-defined, heterogeneously enhancing mass in the distal femoral shaft and meta-diaphysis with extension into the epiphyseal region. The femoral condyle was destroyed and malformed. At the center of the lesion, a large loculated area was observed, which was T2/PD hyperintense and T1 iso- to hyperintense, with restricted diffusion and minimal heterogeneous post-contrast enhancement, consistent with an intramedullary abscess. There was marked surrounding edema, and myositis involving the anterior compartment and adductor muscles. Subtle fluid extended into the surrounding intramuscular planes. Also, a sinus tract was demonstrated laterally adjacent

to the knee joint, and low-grade joint effusion with homogenous thickened synovium was witnessed. The patella showed heterogeneous signal abnormalities with a prominent edema and severe thinning of the lateral cartilage facet. In contrast to these extensive findings, the reconstructed tibia, fibula, and neurovascular structures were intact.

Laboratory analyses during this time revealed initially leukopenia, followed by the leukocytosis associated with localized infection. Hemoglobin levels were low, with a mild decrease in platelet count without the need for transfusion. Blood cultures were negative, and inflammatory markers were mildly elevated, supporting a diagnosis of localized osteomyelitis rather than systemic sepsis. Conservative management was started with intravenous antibiotics, cefuroxime, and then linezolid. To aid with healing, local wound care measures were used, including glyceryl trinitrate ointment. Oral analgesics and laxatives were administered to control pain and gastrointestinal activity. Although a loculated medullary abscess had formed, she exhibited no signs of systemic sepsis, and immediate surgical drainage was not pursued.

After medical stabilization, 3 additional cycles of neoadjuvant chemotherapy were given to maximize tumor control before surgical intervention. Subsequent imaging demonstrated that the tumor was still closely abutted to the articular surface and extended medially and laterally beyond the femoral cortices. The rectus femoris muscle appeared normal but some suspicious involvement of portions of the vastus intermedius muscle was noted while the vastus medialis and vastus lateralis muscles were uninvolved. The neurovascular structures were still preserved.

From the imaging findings and multidisciplinary review, the decision was made to proceed with wide local excision of the distal femur with intraarticular resection approximately 7 cm from the joint line with concomitant resection of involved fibers of the vastus intermedius and portions of the vastus medialis and lateralis muscles. Reconstruction options included limb-salvage surgery with a modular prosthesis or resection arthrodesis with long nail and free vascularized fibular graft placed, the latter of which was ultimately selected for this case. Wide local excision with reconstruction was performed with adequate oncologic margins and limb preservation. The patient was stable postoperatively with no acute complications. Plans were made for the continuation of adjuvant chemotherapy following surgical recovery, along with close follow-up to monitor for signs of recurrence or infection.

Radiograph Right femur lateral projection shows external fixators through the distal thigh with a lytic lesion having a wide zone of transition and internal coarse calcifications resulting in near destruction of the underlying femoral meta-diaphysis as well as epiphysis. Associated with significant surrounding soft tissue.

Coronal T1 pre- and post-contrast images of the same patient after chemotherapy show interval reduction in the size of the heterogeneous mixed signal intensity mass in the distal femoral meta-diaphysis with internal calcifications, associated Codman's triangle, and intramedullary abscess formation. Significant surrounding enhancement was associated, suggesting myositis in the muscles and subcutaneous tissues, thickening and enhancement of the synovium at the knee joint with mild joint effusion.

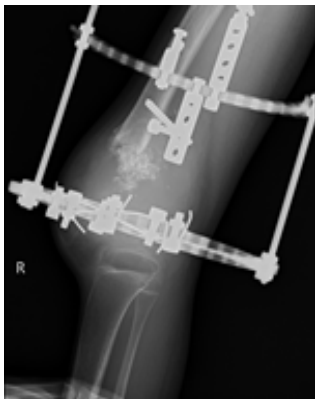

Figure 1: MRI of the right femur lateral projection.

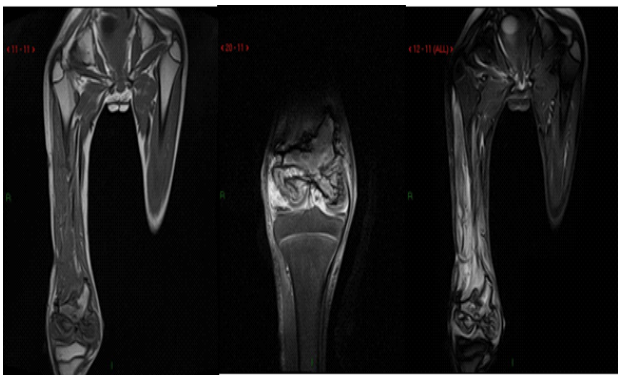

Figure 2: MRI of the right knee comparing pre- and post-operative images.

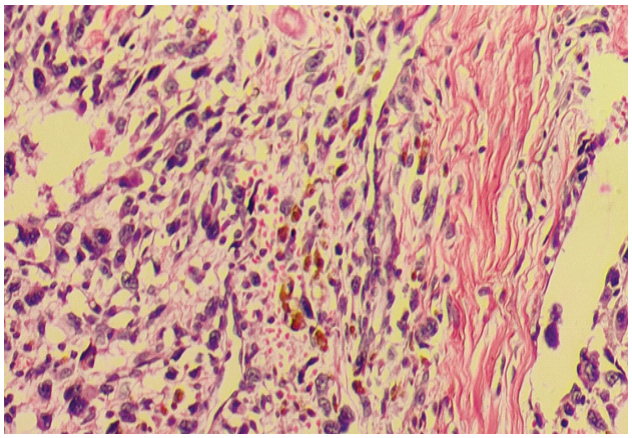

Figure 3: Histopathological section of the distal femoral lesion in the same patient with telangiectatic osteosarcoma.

Microscopic examination reveals multiple blood-filled cystic spaces separated by septa containing highly pleomorphic malignant spindle cells with marked nuclear atypia and brisk mitotic activity. Scattered multinucleated osteoclast-like giant cells are present within the hemorrhagic background.

Discussion

Telangiectatic osteosarcoma (TOS) is a rare histological subtype of osteosarcoma, comprising 2-12% of osteosarcomas in the first two decades of life [1]. Radiologically and histologically, TOS represents a challenge, owing to cystic areas filled with blood that can sometimes be confused with benign lesions such as aneurysmal bone cysts (ABC) [2]. This highly aggressive biological behavior of TOS needs early and accurate diagnosis, as misinterpretation may lead to a delay in definitive therapy and affect prognosis negatively [3].

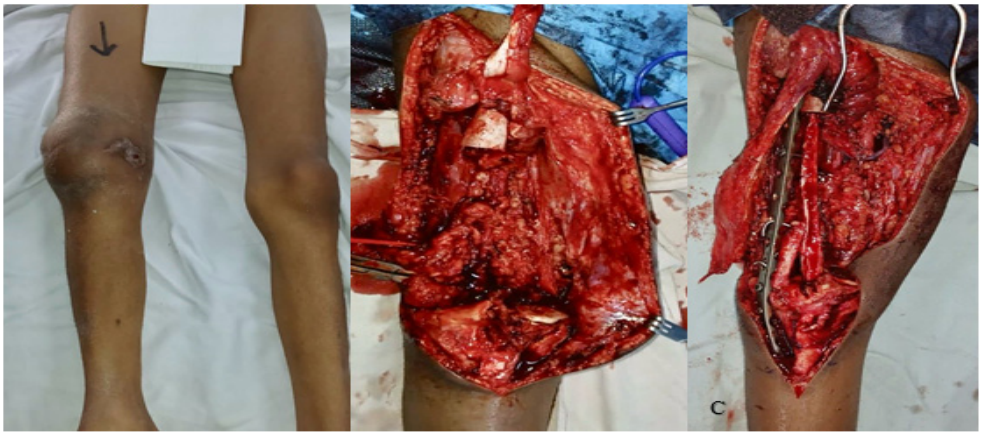

Figure 4: Composite image of the affected limb preoperatively and intraoperatively.
(A) Preoperative photograph showing swelling, skin changes, and surface ulceration over the right distal femur. **(B)** After WLE of the involved femur, intraoperative exposure reveals wide local excision of the distal femur with resected bony margins and adjacent soft tissue dissection in preparation for limb reconstruction. **(C)** Reconstruction of the bone defect with vascularized fibula and reconstruction plate.

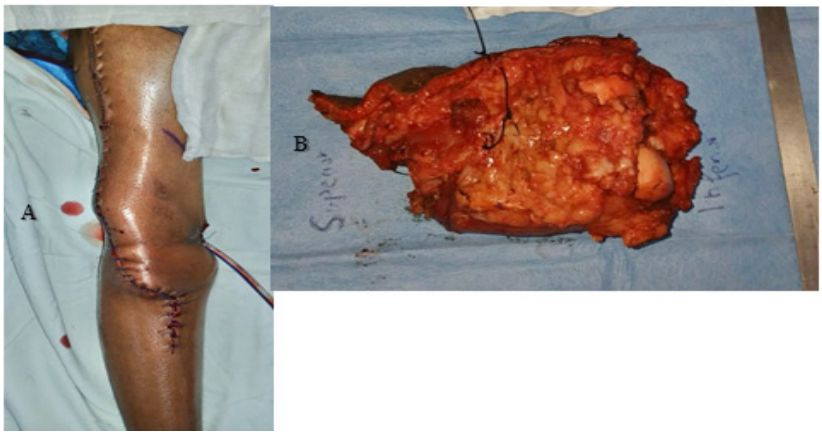

Figure 5: Composite image of postoperative limb status and gross surgical specimen.
(A) Postoperative clinical photograph showing well-aligned reconstructed limb with surgical drains in place. **(B)** Gross specimen of the resected distal femur showing distorted bony architecture and soft tissue involvement.

In the case shown here, preliminary biopsy led to differing histopathological interpretations, complicating the diagnostic pathway. The initial pathological impression based on the presence of atypical spindle cells, pronounced nuclear atypia, and blood-filled cystic areas was telangiectatic osteosarcoma; however, a second opinion favored an aneurysmal bone cyst. Cytological analysis added to the diagnostic conundrum when giant cells and hemorrhagic background were demonstrated, but other malignant features were not defined. Indeed, such diagnostic confusion is not uncommon, given that prior studies have shown underdiagnosis due to sampling genuinely limited biopsy samples, which may not always capture malignant osteoid production [4]. Moreover, ABC and TOS exhibit overlapping radiological characteristics, such as expansile lytic lesions with fluid-fluid levels on MRI, rendering imaging alone inadequate for final distinction [5].

Descriptive correlation of clinical, radiological, and pathological features is still to be determined for diagnosis. TOS is typically more aggressive than ABC, with cortical destruction, periosteal reaction (often with Codman's triangle appearance), and soft tissue invasion [6]. The extensive cortical destruction, pathological fracture, and soft tissue extension observed on imaging were more suggestive of malignancy, necessitating re-evaluation and leading to the final diagnosis of TOS in the present case. The importance of multidisciplinary discussion cannot be overstated, as it was critical in resolving the initial diagnostic uncertainty.

The clinical course was further complicated by superimposed infection, notably osteomyelitis. Follow-up MRI imaging revealed intramedullary abscess formation, myositis, sinus tract development, and mild joint effusion. While infection is an uncommon complication when dealing with malignant bone tumors, it is nonetheless well documented to occur as a complication of surgical interventions, including placement of external fixators or as a result of chemotherapy-induced immunosuppression [7]. Differentiation of infection from tumor progression on imaging may be difficult, as both processes may present with edema, fluid collections, and enhancement. In this case, diffusion-weighted imaging and attention to patterns of enhancement helped to reveal the intramedullary abscess. Intravenous antibiotics were sufficient to control the infection while definitive surgical planning was conducted without significant further delay.

Surgical management in cases of telangiectatic osteosarcoma is focused on the attainment of wide resection margins whilst preserving limb function wherever possible. The choice of limb-salvage versus amputation is influenced by tumor lo-

cation, size, neurovascular invasion, and in response to chemotherapy [8]. In this case, the wide local excision with the intra-articular resection of the distal femur was performed successfully. If surgeons had considered for mobilization of these structures, preservation of critical structures such as the rectus femoris and major neurovascular bundles, and performed resection arthrodesis and/or modular prosthesis, it would have been possible. Amputation was avoided in favor of limb salvage given the preservation of the functional structures and oncologic margins that were within an acceptable range.

The prognosis of telangiectatic osteosarcoma, when identified and treated appropriately, is similar to that of conventional osteosarcoma, with five-year survival rates reported between 55% and 70% [9]. Prognostic factors for better outcomes include early diagnosis, favorable histological response to neoadjuvant chemotherapy, and accomplishment of wide surgical margins. Yet, infection of a primary malignancy is arguably a poor prognostic unknown due to its potential to delay chemotherapy and induce local recurrence [10]. In this case, early detection and aggressive management of infection likely contributed to a favorable clinical course.

Conclusion

This case serves to highlight vital teaching points including the need for direct correlation of pathology and imaging to avoid misdiagnosis, the recognition of telangiectatic osteosarcoma as a potential diagnosis in the differential diagnosis of cystic bone lesions in a child, the complications that can arise when infection is superimposed on malignancy, and the importance of multidisciplinary management to achieve the best possible outcome. Thus, early recognition and aggressive complex multidisciplinary management were paramount in untangling the interactions between malignancy and infection in this patient, leading to ultimately successful limb-salvage surgery and continuation of oncologic therapy.

Declarations

Ethics approval and consent to participate: This case was conducted following ethical standards. Institutional Review Board (IRB) approval was obtained from Shifa International Hospital with the approval number IRB 155-25. Written informed consent was acquired from the patient's legal guardian for the presentation, investigation, and treatment outlined in this report.

Consent for publication: Written informed consent was obtained from the patient's legal guardian for the publication of this case report and accompanying images. All identifying details have been removed to preserve anonymity.

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Authors' contributions:

Wajahat Mirza: Conceptualization, data collection, literature review, manuscript drafting.

Dr. Ishfaq Ahmad: Clinical supervision, oncological diagnosis and treatment, manuscript review.

Dr. Sundus Dadan: Research coordination, manuscript editing, and revision.

Dr. Mamoon Rashid: Surgical planning, operative management, intraoperative decision-making.

Dr. Rabia Tariq: Pediatric oncology input, chemotherapy planning, case review.

Dr. Laiba Masood: Radiological evaluation, imaging interpretation, figure analysis.

Dr. Shahrukh Mohmand: Surgical assistance, intraoperative photography, case documentation.

All authors reviewed and approved the final manuscript.

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