JOURNAL OF CLINICAL CASE STUDIES, REVIEWS & REPORTS
CSK SCIENTIFIC PRESS
Case Report
Volume 4 | issue 5
ISSN: 3069-0781

Parosteal Aneurysmal Bone Cyst Mimicking Telangiectatic Osteosarcoma: Case Report

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Submitted: 29 Sep 2025 **Accepted:** 06 Oct 2025 **Published:** 10 Oct 2025

Citation: *Podberesky, D., Chandra, T., Lacerda, I. R. A., Stanton, R. P., & Miles, L. (2025). Parosteal Aneurysmal Bone Cyst Mimicking Telangiectatic Osteosarcoma: Case Report. J of Clin Case Stu, Reviews & Reports 4(5),1-6.*

Abstract

Parosteal aneurysmal bone cyst is a rare subtype of aneurysmal bone cyst. We report a case of a 15 old year girl with a diagnosis of parosteal aneurysmal bone cyst which mimicked telangiectatic osteosarcoma. The radiological and pathological features of both lesions are discussed with a detailed review of the literature. The aim of this report is to raise the awareness of paraosteal aneurysmal bone cyst as differential diagnosis when dealing with an expansile lytic or cystic lesion in the metaphysis of a long bone, with imaging features mimicking a telangiectatic osteosarcoma. It is important to make an accurate diagnosis since PABC is benign and TOS is malignant, and the treatment and prognosis are totally different. The index case is a good example of these two lesions having significant overlapping features based on imaging, which made tissue examination crucial to render a definitive diagnosis.

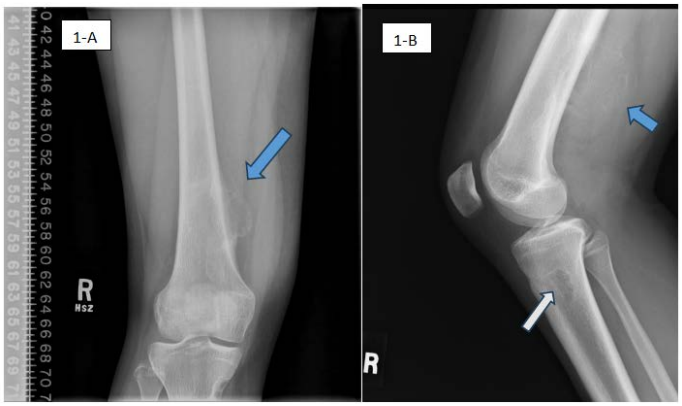
Keywords: Parosteal Aneurysmal Bone Cyst, Aneurysmal Bone Cyst, Telangiectatic Osteosarcoma, Overlap, Mri, Histology.

Abbreviations

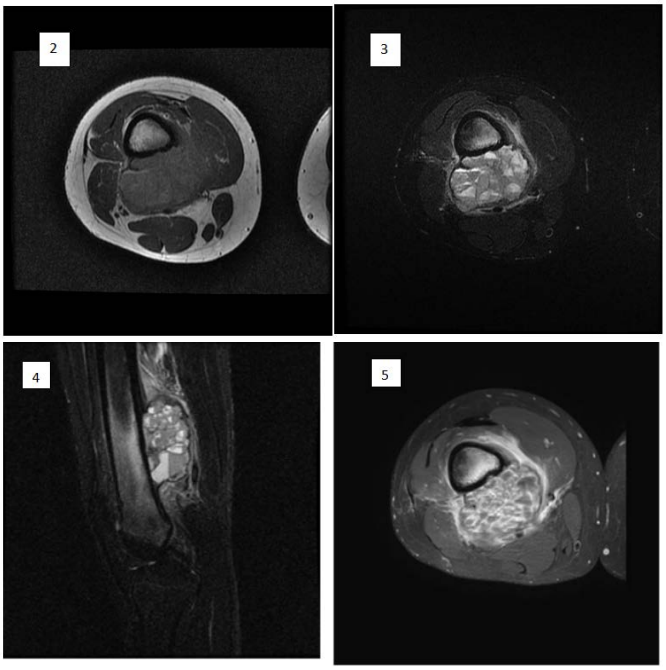
AP = Antero-posterior
MRI = Magnetic resonance imaging
CT = Computed tomography
ABC = Aneurysmal bone cyst
PABC = Parosteal aneurysmal bone cyst
TOS = Telangiectatic osteosarcoma
OS = Osteosarcoma
WHO = World Health Organization

Case Report

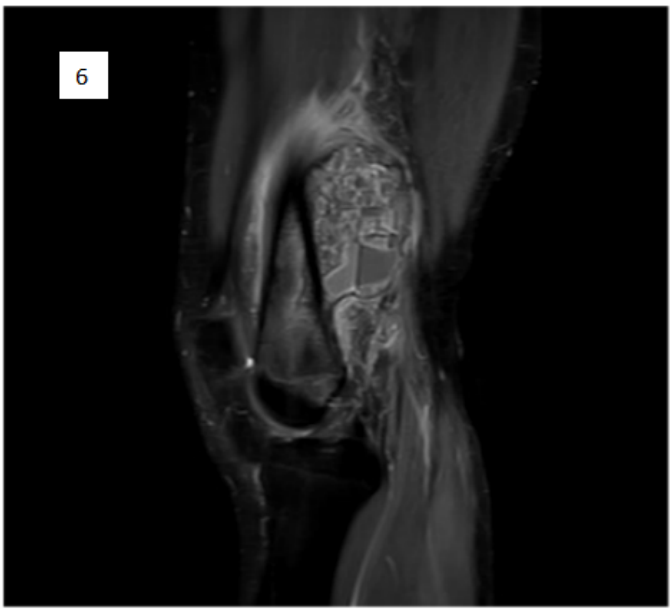
A 15 old year girl was referred to the orthopedic department of our Children's Hospital with 2 months history of leg pain that was thought to be caused by strained muscles around the right knee. Her leg pain was getting worse despite physical therapy. On physical examination, there was an ill-defined fullness behind the distal portion of the right femur toward the popliteal space. Also noted was tenderness toward the medial posterior side of the right knee. The pulse distal to the knee appeared normal. The groin examination did not reveal any prominent lymph nodes. The skin over this area appeared normal. There was no redness, no dilated vessels and circulation in the toes appeared normal. Laboratory exams were within normal limits, including alkaline phosphatase.



AP and lateral radiographs of the extremity showed an aggressive appearing periosteal- based mass along the posteriomedial aspect of the metadiaphysis of distal right femur. The mass has a mildly sclerotic rim and predominantly lucent center with associated aggressive appearing periosteal reaction with lamelled appearance (blue arrow). Incidental note was made of a nonossifying fibroma of the proximal right tibia medially (white arrow) (figure 1-A; 1-B).



MRI of the right lower leg with contrast was performed on a 1.5 T MRI system, with a standard extremity coil. Within the distal right femoral metadiaphysis posteromedially, there was a peripheral, periosteal-based, aggressive, relatively well-defined mass measuring 7.3 x 5.3 x 5.4 cm. The mass had predominantly intermediate signal intensity on T1-weighted images and mixed signal intensity on T2-weighted images. This mass contained innumerable internal small cystic spaces with fluid-fluid levels containing material of varying signal intensity. Associated aggressive appearing periosteal reaction along the medial distal femur was noted. The mass enhanced heterogeneously after contrast administration (Figures 2,3,4,5, and 6).



A 2.1 cm non-ossifying fibroma was noted within the medial aspect of the right proximal tibia metaphysis. No other masses or joint effusions (figures 2,3,4, 5, and 6) were identified.

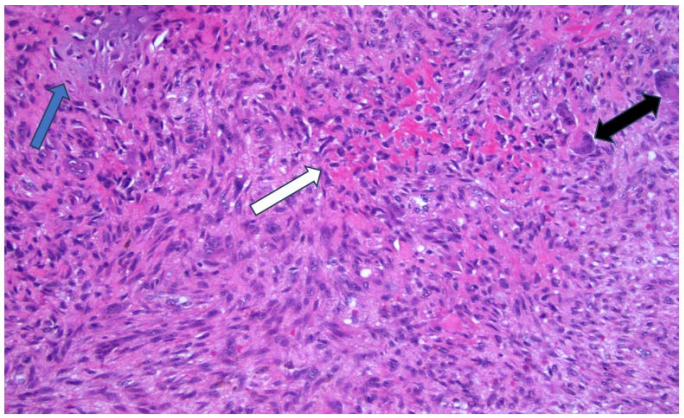


Figure 7 - The fibrous septa is composed of dense cellular proliferation of fibroblasts in the background. Basophilic bone (blue arrow), osteoid with osteoblast rimming (white arrow) and osteoclastic-like giant cells are seen (black arrows).

CT of the chest was normal. No metastatic nodules were noted. The patient underwent an open biopsy of the lesion through a longitudinal incision at the medial aspect of the distal femur. A pie shaped section of the rind was taken and within the rind was a nearly completely blood-filled cavity with a modest amount of soft tissue within the cavity. A section of the rind and a section of the soft tissue within the cavity were sent for frozen section. The frozen section revealed a fibrous lesion but cannot determine whether it is benign or malignant. The diagnosis of ABC was made on the permanent sections.

The resected lesion is a dark red, round, multiloculated cystic mass (figure 7), with numerous septa of variable thickness. The biopsy and resection specimens show similar histologic features. The septa are composed of fibroblastic spindle cells arranged in fascicles. The cellularity is variable and mitotic figures are easily to be found in the more cellular areas. Embedded in the fibroblasts, there are many osteoclast-like giant cells. Basophilic bone ("blue bone") and osteoid formation is seen (figure 8). Blood filling spaces and hemosiderin depositions have also been identified. Although there is osteoid formation and mitoses throughout the tumor, a sarcoma component is not identified and the diagnosis of PABC was confirmed on the resection specimen. In addition, tumor cytogenetic study revealed abnormal karyotype involving rearrangement at chromosome 17p13 that is consistent with USP6 gene rearrangement, commonly seen in ABC.

No additional therapy was recommended due to the final diagnosis of a benign lesion.

Discussion

PABC and TOS are different lesions with significantly different treatment and prognosis, which made the accurate differential diagnosis important. Miss diagnosis may lead to devastating consequences to the patient and family. [1-3].

Radiologically and histologically PABC and TOS can have significant overlapping features which made the differential diag-

nosis of these entities difficult [4-6]. TOS and PABC are uncommon subtypes of osteosarcoma and ABC, respectively. Age of presentation and localization are similar for both diseases. We review the key radiological and pathological diagnostic criteria in the differential diagnosis of PABC and TOS. Also, we compare the characteristic features from our index case with those reported in the literature and discuss the differences.

ABC is a well-known lesion of metaphysis of long bones, especially distal femur, and proximal tibia, but may also involve skull and posterior elements of the spine. PABC is the least prevalent subtype, representing 7% of all ABC and occurs periosteally, expands peripherally ultimately penetrating within the bone [7]. Mirra classified the development of ABC in four phases [8]. The incipient phase is characterized by erosion of the cortex and elevation of the periosteum without reactive osteogenesis, which suggests suspected malignancy. These lesions most commonly present in the primary or second decades of life [9, 10]. Similar features are seen with TOS [11-13].

Many theories have been proposed regarding etiopathogenesis of ABC as vascular, traumatic, or genetic. Recent genetic studies suggest that primary ABC is a true neoplasm rather than a vascular disturbance, with identified chromosomal translocation t (q22; p13) and identification of neoplastic cells exhibiting USP6 or CDH11 rearrangements that was identified in this case [14]. The classic radiographic image appearance of ABC is a radiolucent cystic lesion within the metaphyseal portion of the bone expanding its cortex, which contains fluid-fluid levels with solid component, but remains contained by a thin shell of cortex. These findings are highly suggestive of ABC but are not pathognomonic as they can also be seen in cases of TOS [15, 16]. TOS is one of the 8 subtypes of the OS (1, 2, 6, 9, 11). It accounts for 2 to 12 % of all OS [17-19]. TOS usually occurs in metaphysis of long bones, femur, tibia, and humerus as conventional osteosarcoma but can be found in unusual locations such as skull, mandible, spine, metatarsal, and soft tissue. The characteristic features on x- ray, CT or MRI are a lytic or cystic intra or juxta metaphyseal lesion, fluid-fluid levels, cortical destruction, and soft tissue mass, can be seen in ABC, PABC and TOS [20].

When these tumors are associated with peculiar radiographic characteristics, miss diagnosis is possible and were reporte. There are cases where the first diagnosis of ABC was made but after 2 or 3 relapses, revision of histopathologic features was performed and it was concluded as TOS and vice versa as reported by Matsuno at al, in a large series from the Mayo Clinic, and similarly by some other authors. It is critical that the correct diagnostics of TOS versus PABC be done based in some criteria as claimed by WHO Classification. It includes radiological and histological findings, and gross examination.

1) A predominantly lytic destructive lesion with only minimal lesional sclerosis on X-ray
2) A soft cystic cavity like tumor on gross examination
3) Histologically, single, or multiple cystic dilated spaces containing blood or necrotic tumor cells lined or crossed by septa containing anaplastic sarcoma cells and numerous mitoses. Osteoid formation is scanty and thin between tumor cells.
4) In this case, radiologic findings highly suggested TOS, based on multiple fluid -fluid levels and periosteal reaction along of the distal femur (as described in criteria number 1, on x-ray and MRI. The distinction between TOS and ABC is usually with MRI. ABC has limited thin peripheral septa (2-3mm tick), which are often best seen as enhancing structures that lack nodularity on gadolinium enhanced images. Unlike ABC, in TOS the peripheral septa around the hemorrhagic spaces are thickened, nodular and enhanced with gadolinium. At computed tomography (CT) the presence of osteoid matrix within nodular or septal regions (intraosseous or soft tissue component) is a second distinguishing feature from ABC. TOS is associated with aggressive growth features (cortical destruction and extension to soft tissue). In contrast, ABC causes marked remodeling of bone and cortical thinning, but lacks a true soft-tissue component. However, in our case, there were overlapping features on both X rays and MRI and a clear distinction was not possible based on radiologic findings.

Histologic differentiation of ABC and PABC from TOS is usually not difficult because TOS almost always shows highly anaplastic cells with atypical mitoses. The differential diagnosis may be difficult when the lesion at the para or peri osteo location. These may be intermediate or low grade OS associated with a secondary ABC. Histopathology (criteria number 3) of the lesion demonstrated areas of hyper cellularity, increased mitosis, woven bone formation and cytological atypia in the background of ABC. The diagnosis of PABC was based on lack of high-grade sarcomatous cells around the periphery and septations of the blood space. Blood spaces in TOS are pathologically like ABC, although in PABC the sarcomatous cells are absent. Another point we would like to highlight is that a good biopsy sample should be made in the areas pointed by MRI as the most representative of disease. This decreases the possibility of histologic findings be overlooked by limited sample volume obtained by a core needle biopsy. Bahk and Mirra claim that in a retrospective review of 215 cases of ABC, a deep purple, reticulated, chondroid matrix in the wall was found in some cases, what they called “blue reticulated chondroid-like material (BRC)”. This finding is unique for ABC and not found in TOS, which has a value in discriminating ABC from TOS. This finding was seen in our case as well.

In summary, neither radiologic nor clinical assessment is reliable to definitively distinguish ABC from TOS. Therefore, tissue diagnosis is essential, and the final diagnosis is based on histologic and radiologic criteria defined in the WHO Classification. This case had overlapping radiological findings of PABC and TOS, however the histologic diagnosis of PABC was done after discussion and teamwork. As advocated by Matsuno et al is imperative that the diagnosis being based on histopathologic findings [21, 22].

Teaching Point

PABC can mimic TOS and vice versa. Giving the vast difference in treatment of these entities and the potentially serious consequences of misdiagnosis, it is essential that the Radiologist, Pa-

thologist, and the treating physician to be cognizant of the overlapping features of these entities to reach the accurate diagnosis.

Questions

Question 1: Which of the following answer is false?
Answer choice 1: PABC and TOS can overlap radiologic and histologic features.
Answer choice 2: TOS and PABC are uncommon subtypes of osteosarcoma and ABC, respectively.
Answer choice 3 (applies): PABC and TOS are found in different decades of life. It is a clue for the differential diagnosis.
Answer choice 4: PABC and TOS are more frequently found as expansible lytic or cystic lesion in the metaphysis of a long bones.
Answer choice 5: ABC as found as a true neoplasm.

Explanation:
1.PABC and TOS can overlap radiologic and histologic features. [Radiologically and histologically PABC and TOS can have significant overlapping features which made the differential diagnosis of these entities difficult.]
2.TOS and PABC are uncommon subtypes of OS and ABC, respectively. [TOS is one of the 8 subtypes of the OS. It accounts for 2 to 12 % of all OS. PABC is the least prevalent subtype, representing 7% of all ABC and occurs periosteally, expands peripherally ultimately penetrating within the bone.]
3.PABC and TOS are found in different decades of life and different localization. It is a clue for the differential diagnosis. [Age of presentation and localization are similar for both diseases.]
4.PABC and TOS are more frequently found as expansible lytic or cystic lesion in the metaphysis of a long bones. [The classic radiographic image appearance of ABC is a radiolucent cystic lesion within the metaphyseal portion of the bone expanding its cortex, which contains fluid-fluid levels with solid component, but remains contained by a thin shell of cortex. These findings are highly suggestive of ABC but are not pathognomonic as they can also be seen in cases of TOS.]
5.ABC as found as a true neoplasm. [Recent genetic studies suggest that primary ABC is a true neoplasm rather than a vascular disturbance, with identified chromosomal translocation t (16; 17) (q22; p13) and identification of neoplastic cells exhibiting USP6 or CDH11 rearrangements (18, 19, that was identified in this case.]

Question 2: Which of following answer is false?
Answer Choice 1: MRI is an important complementary exam to differentiate PABC from TOS.
Answer Choice 2: ABC, PABC and TOS, can overlap characteristics on X-ray, CT and MRI.
Answer Choice 3: Cystic lesion and fluid-fluid levels are common radiologic findings seen in PABC and TOS.
Answer Choice 4 (applies): ABC is exclusively found in metaphysis of long bones.
Answer Choice 5: TOS is found in metaphysis of long bone, but also in other sites.
Explanation:
1.MRI is an important complementary exam to differentiate PABC from TOS. [The distinction between TOS and ABC is usually with MRI. ABC has limited thin peripheral septa (2-3mm tick), which are often best seen as enhancing structures that lack nodularity on gadolinium enhanced images.]
2.ABC, PABC and TOS, can overlap characteristics on X-ray, CT and MRI. [The characteristic feature on x- ray, CT or MRI are a lytic or cystic intra or juxta metaphyseal lesion, fluid-fluid levels, cortical destruction and soft tissue mass, can be seen in ABC, PABC and TOS.]
3.Cystic lesion and fluid-fluid levels are common radiologic findings seen in PABC and TOS. [The classic radiographic image appearance of ABC is a radiolucent cystic lesion within the metaphyseal portion of the bone expanding its cortex, which contains fluid-fluid levels with solid component, but remains contained by a thin shell of cortex. These findings are highly suggestive of ABC but are not pathognomonic as they can also be seen in cases of TOS.
4.ABC is exclusively found in metaphysis of long bones. [ABC is a well-known lesion of metaphysis of long bones, especially distal femur and proximal tibia, but may also involve skull and posterior elements of the spine.]
5.TOS is found in metaphysis of long bone, but also in other sites. [TOS usually occurs in metaphysis of long bones, femur, tibia and humerus as conventional osteosarcoma (13) but can be found in unusual locations such as skull, mandible, spine, metatarsal, and soft tissue.]

Question 3: Choose the true answer:
Answer Choice 1: The treatment and prognosis for PABC and TOS is the same.
Answer Choice 2 (applies): PABC and TOS overlap features which can lead to a misdiagnosis.
Answer Choice 3: The main criteria for a diagnosis of PABC and TOS is clinical and radiological findings.
Answer Choice 4: TOS is not associated with aggressive growth features.
Answer Choice 5: WHO classification includes radiologic findings as diagnostic criteria only.

Explanation:
1.The treatment and prognosis for PABC and TOS is the same. [PABC and TOS are different lesions with significantly different treatment and prognosis, which made the accurate differential diagnosis important.]
2.PABC and TOS overlap features which can lead to a misdiagnosis. [When these tumors are associated with peculiar radiographic characteristics misdiagnosis is possible and were reported.]
3.The main criteria for a diagnosis of PABC and TOS is clinical and radiological findings. [Neither radiologic nor clinical assessment is reliable to definitively distinguish ABC from TOS. As advocated by Matsuno et al (1) is imperative that the diagnosis being based on histopathologic findings.]
4.TOS is not associated with aggressive growth features. [TOS is associated with aggressive growth features (cortical destruction and extension to soft tissue).]
5.WHO classification includes only radiologic findings as diag-

nostic criteria. []). It is critical that the correct diagnostics of TOS versus PABC be done based in some criteria as claimed by WHO Classification. It includes radiological and histological findings, and gross examination.]

Question 4: Which of following answer is true?
Answer choice 1 (applies): The histologic differential diagnosis between PABC and TOS is based on a lack of sarcomatous cells.
Answer choice 2: "Blue reticulated chondroid-like material (BRC)" is a unique finding for TOS on the histologic diagnostic.
Answer Choice 3: Blood spaces in TOS are pathologically different to ABC, which helps in differentiate these two lesions.
Answer choice 4: Sometimes TOS does not show anaplasic cells, that brings difficulty to differentiate TOS from PABC.
Answer choice 5: Para or periosteal location and background of ABC can facilitate the diagnosis between PABC and TOS.

Explanation:
1.The histologic differential diagnosis between PABC and TOS is based on a lack of sarcomatous cells. [The diagnosis of PABC was based on lack of high grade sarcomatous cells around the periphery and septations of the blood space.]
2."Blue reticulated chondroid-like material (BRC)" is a unique finding for TOS on the histologic diagnostic. [Bahk and Mirra (19) claim that in a retrospective review of 215 cases of ABC, a deep purple, reticulated, chondroid matrix in the wall was found in some cases, what they called "blue reticulated chondroid-like material (BRC)". This finding is unique for ABC and not found in TOS, which has a value in discriminating ABC from TOS.]
3.Blood spaces in TOS are pathologically different to ABC, which helps in differentiate these two lesions. [Blood spaces in TOS are pathologically similar to ABC, although in PABC the sarcomatous cells are absent.]
4.Sometimes TOS does not show anaplasic cells. [Histologic differentiation of ABC and PABC from TOS is usually not difficult because TOS almost always shows highly anaplastic cells with atypical mitoses.]
5.Para or periosteal location and background of ABC can facilitate the diagnosis between PABC and TOS. [The differential diagnosis may be difficult when the lesion at the para or peri osteo location. These may be intermediate or low grade OS associated with a secondary ABC.]

Question 5: Which of following answer is false?
Answer Choice 1: Although PABC is a para or periosteal lesion it can penetrate the bone.
Answer Choice 2 (applies): On MRI TOS and ABC have the same findings: the peripheral septa around the hemorrhagic spaces are thickened, nodular and enhance with gadolinium.
Answer Choice 3: ABC lacks a true soft tissue component.
Answer choice 4: The classical radiologic finding of ABC is expansive cystic lesion with fluid-fluid level, with a thin cortical.
Answer choice 5: ABC has many etiopathogenic theories.

Explanation:
1.Although PABC is a periosteal lesion it can penetrate de bone. [PABC is the least prevalent subtype, representing 7% of all ABC occurs periosteally, expands peripherally ultimately penetrating within the bone.]
2.On MRI TOS and ABC have the same findings: the peripheral septa around the hemorrhagic spaces are thickened, nodular and enhance with gadolinium. [Unlike ABC, in TOS the peripheral septa around the hemorrhagic spaces are thickened, nodular and enhance with gadolinium.]
3.ABC lacks of true soft tissue component. [In contrast, ABC causes marked remodeling of bone and cortical thinning, but lacks a true soft-tissue component.]
4.The classical radiologic finding of ABC is expansive cystic lesion with fluid-fluid level, with a thin cortical. [The classic radiographic image appearance of ABC is a radiolucent cystic lesion within the metaphyseal portion of the bone expanding its cortex, which contains fluid-fluid levels with solid component, but remains contained by a thin shell of cortex.]
5.ABC has many etiopathogenic theories. [Many theories have been proposed regarding etiopathogenesis of ABC as vascular, traumatic or genetic.

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