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Case Report

Periosteal Aneurysmatic Bone Cyst: A Rare Case Treated by Pre-Surgery Endovascular Embolization

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Abstract

In this work we report the rare case of a periosteal aneurysmatic cyst found in a 30-year-old male patient. The patient came to our hospital for the detection of a painful swelling located on the lateral side of the right thigh; for this reason, he underwent objective examination and then X-rays, MRI examination and PET-CT. Once the diagnosis was established, in agreement with the orthopedists, in view of the rich vascularization of the lesion, it was decided to perform a pre-operative arterial embolization in order to reduce the complications related to the intervention, specifically reducing the risk of intraoperative bleeding, and promoting a better post-operative course. In this text we report the images and the technique used to perform the preoperative arterial embolization and therefore the results obtained.

Keywords: Periosteal Aneurysmatic Bone Cyst; Transcatheter Embolization; Interventional Radiology; Combined Treatment

Introduction

Aneurysmal bone cyst (ABC) is a relatively rare benign, locally destructive tumor of the bone. It occurs as a primary bone cyst in ~79% of cases, but it can be also a secondary lesion arising from other osseous conditions in ~20% of case. It represents ~1% of all bone tumors. Most ABC has been reported to occur in the first three decades of life (~95%) with the peak age of onset < 20 years [1].

About its pathogenetic origin there are conflicting articles with regard whether ABCs represent a neoplastic or a reactive process. In the past ABCs were originally thought to be reactive lesions caused by venous hypertension leading to vascular dilatation. However, several recent studies have documented that ABCs are not correlated to venous hypertension condition and then they are neoplastic lesions in nature.

This theory is confirmed by genetic studies, that proved a recurrent genetic translocation involving the USP6 gene on

chromosome 17p13 is associated with many primary ABCs. Other less frequent translocations are associated with ABCs [2].

ABC can be localized in any bone however these tumors predominantly occurs in the metaphysis of long bones (65%), the pelvis (12%) and the arch of the spine (12%).

In a recent study Capanna., *et al*. classified ABCs by 5 types (I-V) based on morphological characteristics on plain-X-rays. Type I consist of a central metaphyseal ABCs, type II consists of central ABCs involving the entire segment of bone, type III consist of eccentric metaphyseal ABCs, type IV consist of surface (subperiosteal) ABCs, and type V consist of meta-diaphyseal ABCs [3].

The differential diagnosis may include other benign tumor such as giant cell tumor (GCT), giant cell reparative granuloma (GCRG) and hyperparathyroidism-related Brown tumor [1].

Different treatment options are reported in literature for patients with ABC, some of these are autogenous bone grafting, cementation, or resection of the lesion. Recurrence is not uncommon, ranging from 10 to 50% [1].

The present study reports a rare case of ABC raised from the periosteum of the diaphyseal of the femoral bone (Type IV sec, Capanna., *et al*.). Only a few cases of periosteal ABC have been reported in literature.

Case Report

A 30-year-old male patient presented to our Hospital with a sudden appearance of pain in his right leg while he was working. The pain was acute in onset associated with swelling which gradually increased in size. There was no history of recent trauma, no history of loss of weight. No relevant medical history was reported.

The pain increased with ambulation and it was radiated to the hip, it did not response to common nonsteroidal anti-inflammatory drugs.

On local examination, there was a localized swelling over the posterior region of the right thigh which was tender. No skin changes, no neurovascular deficits on the ipsilateral lower limb was observed.

The patient was admitted to radiology department to make a plain-X-rays. Radiograph demonstrated an expansive bone mass in the posterior diaphyseal of the right femur characterized by sharp margins, cortical erosion phenomenon, without any periosteal shell. Basing on these findings the patient underwent MRI scan that confirmed a rounded-shape bone lesion localized in the diaphyseal of the right femur extending along the posterior and lateral surface of the bone. In axial acquisitions the lesion was localized in externally to the cortex and involves the smooth tissue around the femur; it showed a heterogeneous signal both in T1 and T2 sequences with a septations and polycystic appearance associated with blood-filled spaces with fluid-fluid levels. After ev gadolinium administration there was an enhancement of lesion wall and septations. The lesion also determined erosion of the outer cortex of the femur and oedema both of perilesional soft tissue and bone marrow. The MRI scan in consideration of characteristic polycystic aspect with fluid-fluid levels suggested the diagnosis of ABC although its localization was atypical (Figure 1).

Figure 1a-1d: 1a and 1b represent preoperative anteroposterior and lateral radiographs of the right femur. Lateral view shows an oval cortical erosion in the posterior diaphyseal region with indefinite borders between the bone and the surrounding soft tissue, without any periosteal shell. Axial and coronal STIR sequences (1c and 1d) demonstrate a large multi-locular cystic lesion in subperiosteal location, exceeding in the soft tissues with peri-tumoral soft tissue edema. Note fluid-fluid levels and reactive bone marrow edema.

However according to the rapid onset presentation and the unusual location, the patient underwent to 18F-FDG PET/TC that revealed an increased uptake only of the area corresponding of the notice lesion without other areas of increased uptake.

CT-guided core-needle biopsy was also performed confirming ABC diagnosis; then excision of the lesion was planned.

Considering the massive vascularization demonstrate in the MRI scan we performed a preoperative trans-arterial embolization of the lesion in order to reduce the blood loss during surgery (Figure 2).

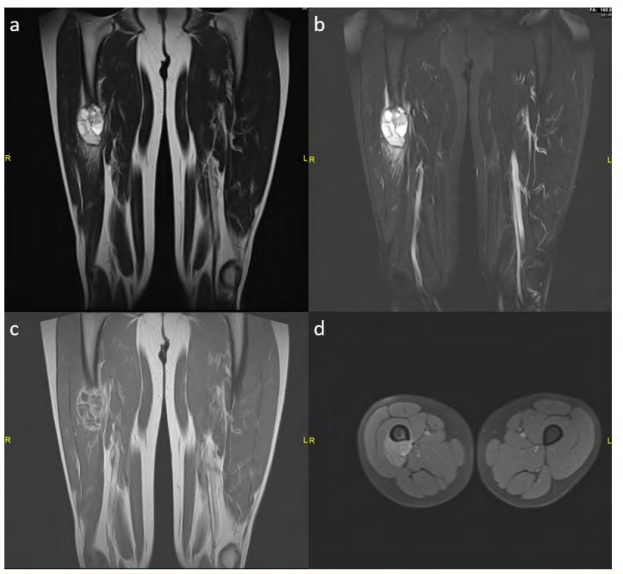

Figure 2: MRI features of lesion: multi-locular cystic lesion with peri-tumoral soft tissue edema in T2 and T2 STIR (Figure 2a and 2b); post-contrast peripheral enhancement with periosteal location in T1 gad+ (Figure 2c) and T1 FS gad+ (Figure 2d).

After local anesthesia, US-guided mono-parietal puncture of the common left femoral artery was performed, and a 5F side-armed sheath introducer was positioned.

An angiographic 5F catheter Simmons shaped was brought up to right common femoral artery and angiography was performed.

The angiography showed a massive vascularity of the lesion with arterial feeders from branches of the right deep femoral artery. Then the catheter was brought up to the right deep femoral artery and arteriography confirmed the lesion vascularity. A microcatheter and a microwire were used to navigate the major arterial feeders then an embolization using a microsphere of 400 um was performed.

Completion arteriography demonstrate stop of flow with correct embolization of the lesion without embolization of no-target tissue (Figure 3).

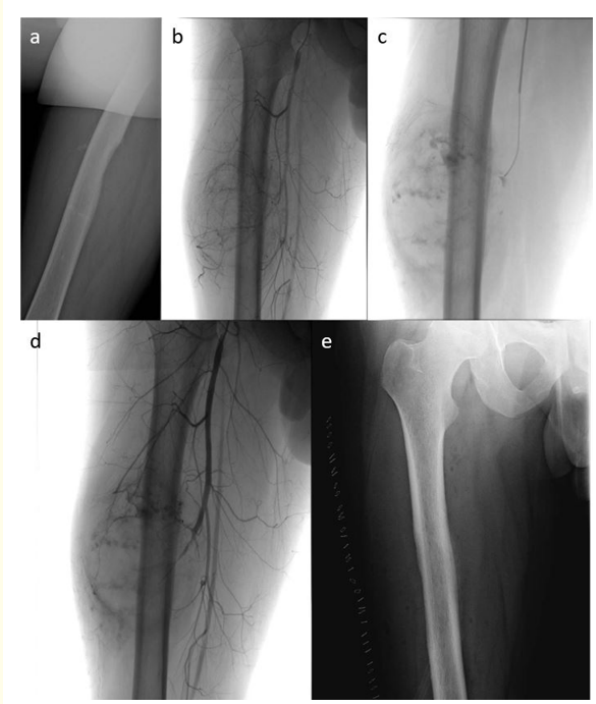

Figure 3a-3e: 3a shows pre-operative radiograph, then 3b-3d are intraoperative radiograph demonstrating respectively pre-embolization angiography, embolization with microspheres and completion angiography that shows complete embolization of the lesion. 3e shows post-surgery radiograph.

At surgery, lateral side of the thigh approach was done, the lesion was dissected from quadricep muscle and removed from the cortex. Deep cortex curettage and phenol/alcohol instillation was

performed as an adjuvant reducing risk of recurrences. Postoperative complications were not noticed and the transcatheter embolization was efficacy reducing the intraoperative bleeding.

At the histological analysis cortical erosion was confirmed but no involvement of medulla was observed. Two days after surgical resection the patient underwent to an internal fixation with carbon intramedullary nail in order to prevent pathological fracture.

Discussion

Periosteal ABCs are very rare bone lesions, in literature only fifty cases were reported, and then often its diagnosis is not easy. A large amount of bone lesions should be included in differential diagnosis, including benign and malignant conditions, both primitive and repetitive, such as ossifying subperiosteal hematoma, periosteal chondroma, periosteal ganglioma, subperiosteal giant cell reparative granuloma, osteoid osteoma, intracortical hemangioma, post traumatic cyst, Ewing's sarcoma, periosteal osteosarcoma, low-grade intracortical osteosarcoma, high-grade surface osteosarcoma, telangiectatic osteosarcoma and periosteal metastatic lesions.

Radiology exams permit to guide the diagnostic process: radiographs are usually the first assessment; they can show a bone lesion with well-circumscribed borders with or without peripheral calcification. Second level imaging is necessary to distinguish the lesion, particularly CT scans can show cortical bone erosion or remodeling and abnormality of the soft tissue around the bone; while MRI can shows typical hyperintense signal in T2 sequencies according to cystic lesion with intralesional septa and also fluid-fluid levels in T1 sequences with hyperintense signal according to hematic part. MRI can also show intensity alteration of soft tissue around the bone lesion according to reactive phenomenon. Both CT and MR scans can be performed with contrast media administration that can reveal the vascularity of the lesion.

Conclusion

The gold standard of the treatment is surgery (i.e. curettage, excision, or resection). Pre-surgery transcatheter embolization of ABCs can be a safe and useful procedure to reduce surgery related bleeding and then improving the outcome.

Disclosure

The authors declare that they have no conflict of interest.

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