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Research article

ANEURYSMAL BONE CYST-SOLID TYPE OF THE CALCANEAL: FROM DIAGNOSIS TO CORRECT MANAGMENT. CASE REPORT AND REVIEW OF LITERATURE

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Abstract

Aneurysmal bone cyst (ABC) is considered a psedotumoral lesion, characterized by single or multi-cavities containing blood. ABC is a rare lesion, accounting for 4% of all bone neoplasms, with an average age at diagnosis of 23 years and more often afflicting females (M/F 1:1.5), generally, ABC is primitive idiopatic of unknown origin, even though it can present following a fracture or develop on a pre-existing lesion such as an osteoblastoma or condroblastoma. Here we discuss a patient who arrived to our hospital with a painful foot which was diagnosed as aneurysmal bone cyst-solid (ABC-solid) located at the calcaneum. Specifically, the management regarding therapy for pain as well as for prevention of fracture due to stress with a particular attention paid to the role of scientific literature.

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Key Words: Aneurysmal Bone Cyst-Solid; Cement, Bone Tumor, Calcaneous.

Introduction

The term aneurysmal bone cyst-type solid (ABC-solid) was first described by Sanerkin et.al, as a non-cystic bone lesion having histological characteristics in the solid parts of a conventional aneurysmal bone cyst[1]. ABC-type solid commonly afflicts the axial skeleton and the short tubular bones of both the hands and feet, whereas it is rarely observed in long bones.
ABC-type solidi is a rare lesion, accounting for 3% of all tumours, that afflicts young patients. The classic variant typically afflicts long bones, almost exclusively metaphyses (80% of cases), while short bones including the vertebrates and pelvis are afflicted in less than 25%. The solid type, first described by Sanerkin, is characterized by a lack of vascular lacune with fibroblastic and histofibroblastic tissue. This variant is known to be more aggressive from a radiographic perspective .Its location is similar to that of the classic form, in that short bones including the vertebrates and pelvis are afflicted in less than 25% of cases.
Below we report on a case of ABC-type solid of the calcaneal bone, treated with curettage and filling of the treated area with cement and therein after periodically controlled with clinical and radiological exams.

Case presentation

A 35 year old male arrived to our emergency room who at recent anamnesis referred only an ongoing pain of the left anterior calcanear region resulting in an evident left-sided limp. As well the patient referred a sudden onset of difficulty in walking over the prior month that was not subsequent to trauma. Regarding remote anamnesis, nothing relevant was referred. A subsequent radiographic exam evidenced a multi-loculated osteolytic lesion measuring 4x3.2 cm at the anterior calcaneous apophysis. A radiograph evidenced an osteolytic lesion measuring 4 x 3.2 cm located apophysis the anterior calcaneum (Fig. 1)



Fig.1 *Radiograph of the left foot.*

A further diagnostic investigation of the skeletal segment using CT was performed

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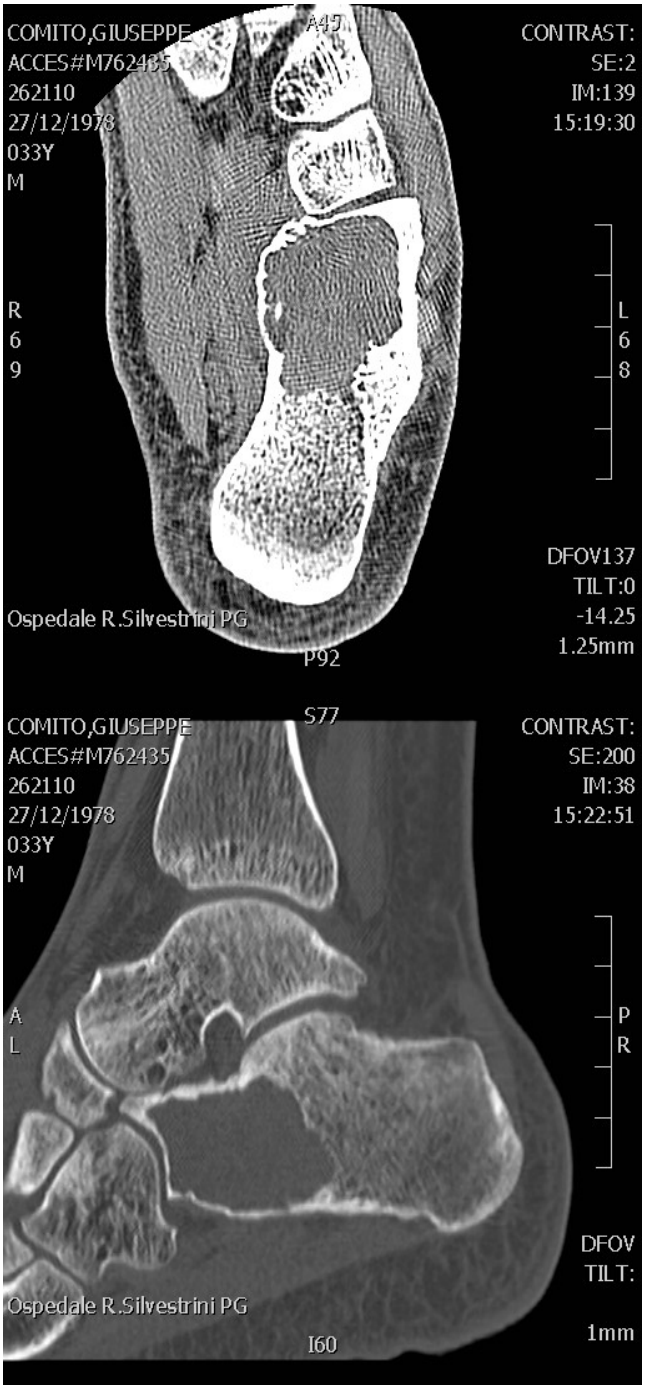


Fig.2 *Axial and crown CT slices showing a large well-delineated osteolytic lesion (circa 4.3 x 5.3cm) of the 'apophysis anterior the calcaneum..*

CT confirmed the presence of a large osteolytic lesion (circa 4.3 x 5.3cm) located apophysis the anterior calcaneum, with well-delineated borders.The cortical bone was at several points markedly thin, the cortical border of spongiosis bone is well defined, but without deliminating sclerotic borders, which are typical of benign tumors .The patient was advised to walk without a load on the left with the aid of crutches, analgesics when needed, a control NMR for the left foot and an bone scintigraphy to assess the infiltration of soft tissues along with metabolic activity of the lesion. The patient returned to our department after having undergone diagnostic exams.

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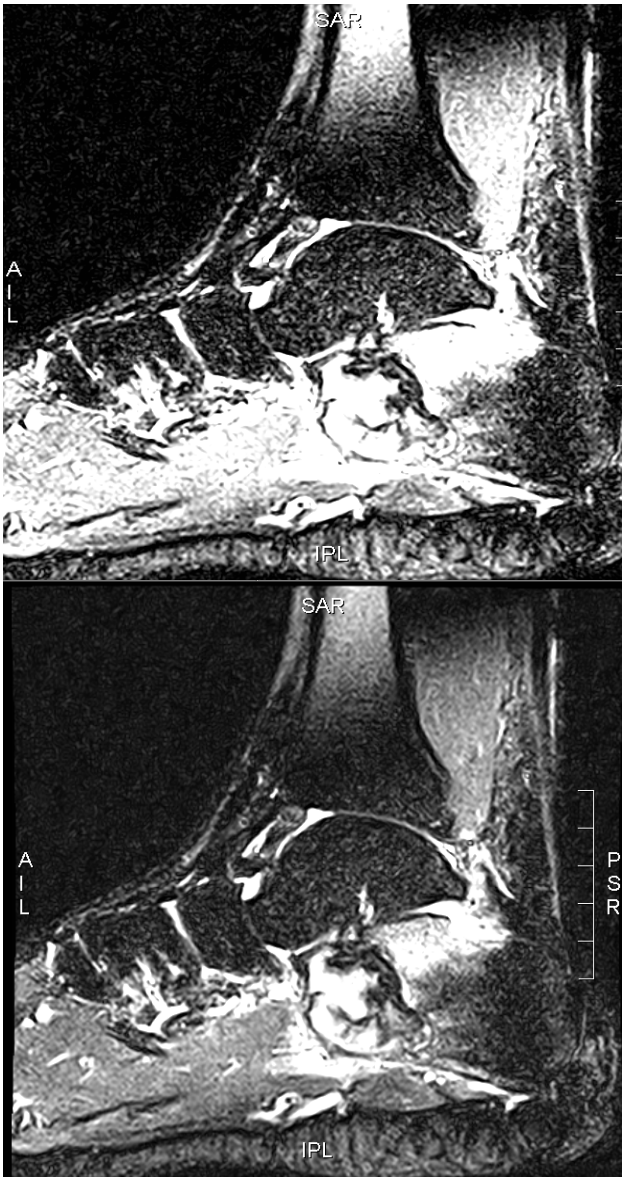


Fig. 3 *NMR shows a focal lesion- third anterior to the calcaneum.*

MRI confirmed the presence of a focal lesion measuring 32x35mm dis-homogeneously hypointense in T1 in sequences T2 dependent localized at the third anterior of the calcaneal involving the anterior articular surface, the anterior calcaneous process and a large part of the sustentaculum. This formation determined a swelling of the osseous cortical that appeared interrupted for a short distance at the height of the medial-anterior margins having a hyperintense aspect in T2 for the surrounding soft tissues and the spongiosis of the adjacent calcaneous lesion.

The exam was performed with and without the endovenous administration of paramagnetic MDC. Following this, the hyperintense portion of the lesion, the osseous spongiosis and the adjacent soft tissues, particularly on the plantar side. The MRI suggested primitive osseous neoformations of the calcaneal The segmented bone scintigraphy (Fig.4) of the feet executed with 99mmCT-HDP and with trifascia technique evidenced hypercaptation of the radiopharmaco in the early vascular phase at the left calcaneal. At the subsequent late static revelations there persists hyperfixations of the tracer at the left calcaneal. The study concluded with a whole body acquisition that did not evidence ulterior fixations of the drug having pathological significance.

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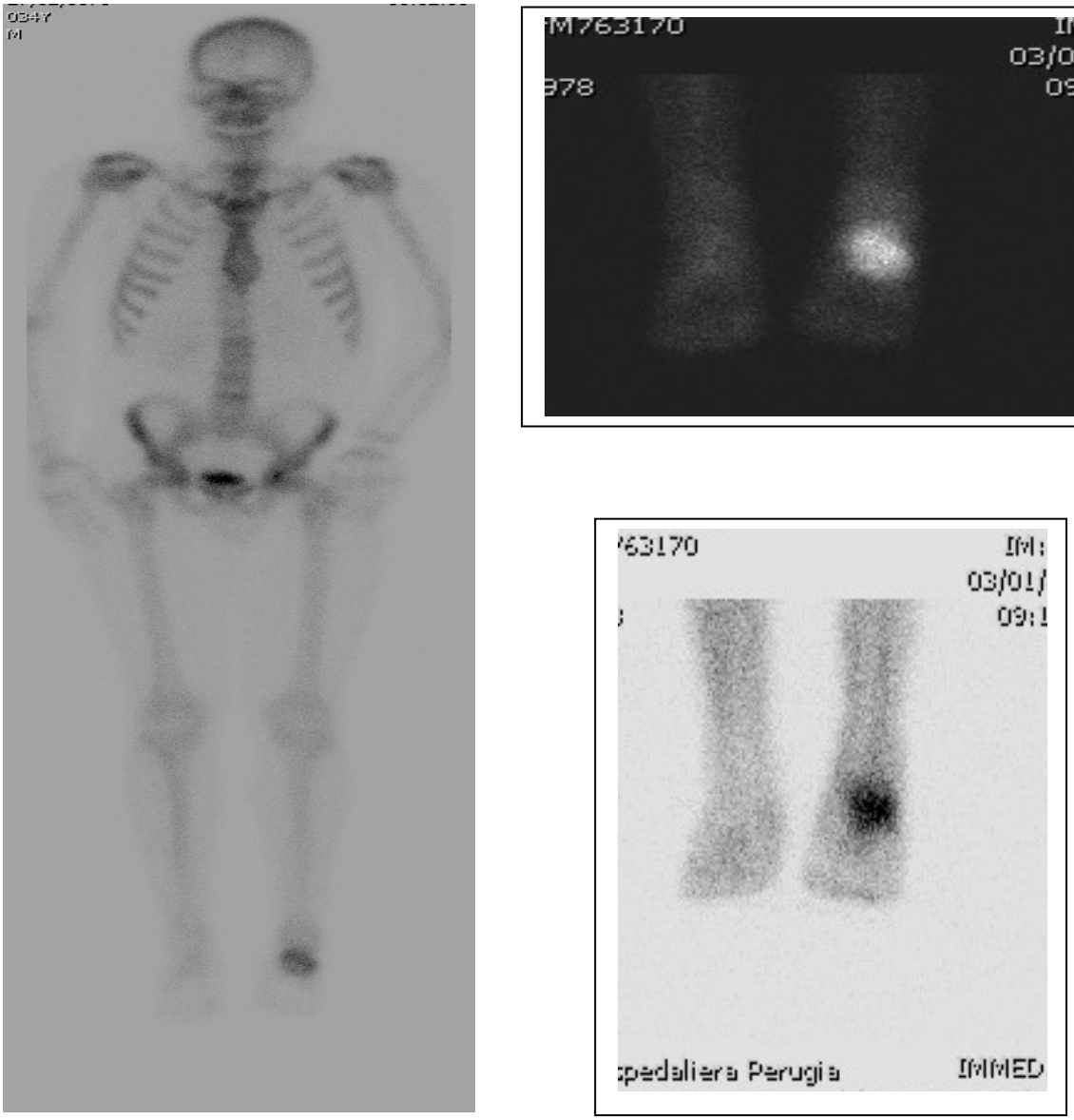


Fig. 4 *Hypercatation at the calcaneal, sign of an active lesion*

The scintigraphy suggests osseous lesions of the left calcaneal having an elevated activity and for this, a guided CT bone biopsy was recommended. The incisional bone biopsy evidenced a proliferation of cells lacking atypical form ,with focal hemosideriniche deposits and numerous giant osteolytic-similar cells . Findings are compatible with a diagnosis of aneurysmal bone cyst-solid type (Fig. 5).

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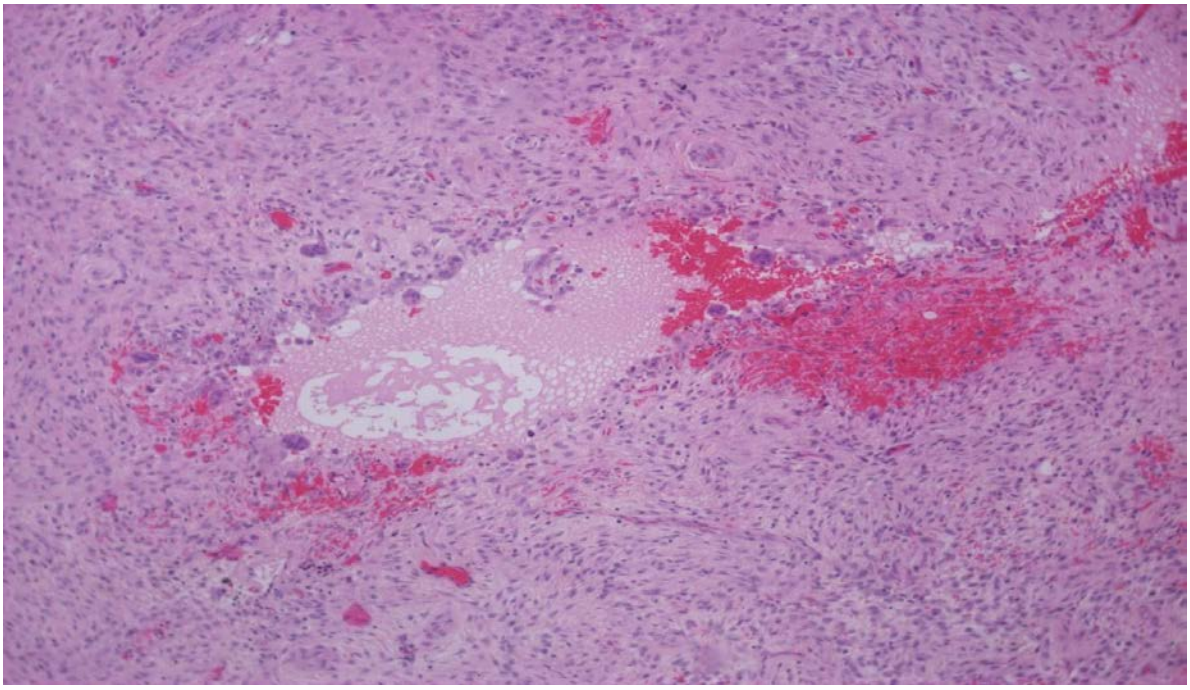


Fig. 5 *Small aneurysmal cystic spaces and fibrous walls with fused cells and giant multinucleated cells. Ematossilina-eosin colored ; enlarged x40.*

Curettage and burrage were performed as well as a refilling with cement, post-surgery the patient walked for 2 months with the aid of crutches, analgesic therapy as needed and periodic control radiographs to exclude for the onset of algodystrophy. A control MRI circa 1 yr from surgery(Fig. 6)

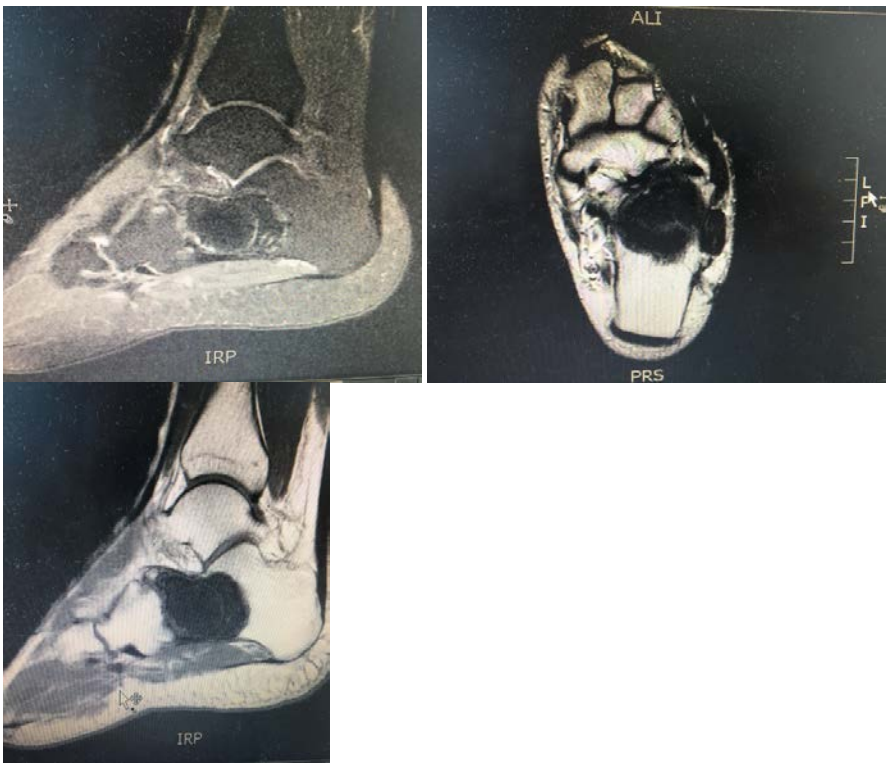


Fig. 6 *Control MRI 1 year after surgery. Curettage of the lesion is evidenced subsequently filled with cement, axial and coronal slices in T1 and T2*

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The patient has returned to his normal every-day activities with a resolution of symptomology. While, due to his occupation as a farmer he has referred episodes of plantar fascia which are resolved by physical therapy as well as functional rest. After three years of follow up there have not been any episodes of recurrence: The patient continues to undergo annually a control MRI and will do so up until the fifth post-operative year as recommended by guidelines.

Discussion

Aneurysmal cystic lesions is a rare pseudo-tumoral lesion of a probable reactive nature that interests the metaphysis or meta-diasphysis in 58% of cases that has sharing of neo-produced periostal material in 66% of cases, responsible for the radiological sign of a border on the bone surface, which is useful for diagnosis[2]. The most frequently cited clinical characteristics of solid aneurysmal cysts include pain, swelling. limping, sedentary habit due to pain during physical activity as well as secondary signs including pathological fractures. A previous trauma is reported in less than 25% of cases[3]. The solid type differs from the classic type for radiological and histological characteristics ;in that vascular lacunar characteristics are either minimally present or lacking. This is so, even if reticolare lasso tissue, fibroblastic and histiofibroblastic typical of ABC[4]. The solid variant rarely or only in its initial phases shows its benign nature from radiograph. Indeed, it instead appears to be a osteolytic lesion with blurred borders having scares or no reaction on the adjacent bone. On radiograph the lesions have a central localisation in 23% of cases and an eccentric location in 58% of cases while in 19% of cases they are positioned on the surface of the bone cortex[5]. It afflicts the short bones in 20% of cases, mostly the vertebrae, but rarely only the vertebral body, very rarely is it found in the calcaneum (4%) often in the anterior region[6]. In accordance with the staging of benign tumors from a radiographic perspective, we can divide ABC-solid into 1)quiescent,2)active or 3)aggressive.
In the quiescent stage, where radiographic characteristics do not allow for a distinction form the classic form, an osteolytic area is present which has a thinning of the cortical along with a transition zone that is well defined without sclerosis and distension of the soft tissues.
The active stage, similar to the first, evidences incomplete cortical margins as well as an ample transition zone.
In the aggressive stage, the osteolysis causes an expansion of the cortical, if not its destruction, with marginal sclerosis on the endostal side of the cavity and early invasion of the adjacent soft tissues. In several cases there is an increase in the periostosis, a gauge of the sizeable local aggression of the lesion. Ultrasounds are always used as a imaging technique of irst approach in order to evaluate the surface of the masses, to recognise lesions that originate from bone structure of the lesion of soft tissues, to individualise the mobile fluid-fluid levels that are characteristic of aneurysmal cysts and finally carry out eco-guided bioptical procedures for both an accurate diagnosis and an appropriate treatment[7]. CT and MRI are essential for better defining the lesion, allowing in some cases to formulate a differential diagnosis. With these methods a better evidencing of the involved soft tissues and the reaction of the periostosis can be obtained. As well, these methods allow to investigate regions with complex bone anatomies which with conventional radiograph difficult to explore. With CT it is possible to evidence a hyperdense line on the exterior border of the lesion which is “cortex shell”

An interuption of the cortical can be observed at the same time, even though the periostitis remains whole. MRI has proven to be a complimentary to CT for its capacity to estimate the rapport of the cystic masses with adjacent tendon structures, assessing therein for its extension.
ABC-classic appears on MRI as an expansive lesion with well-defined margins and the presence of septum that deliminate the cystic cavity. A characteristic sign, though not pathognomic, is the endocavity fluid levels generated by the sedementation of the coagulated components compared to that serous. MRI evidences a homogeneous hypo-intensity of the signals in weighted T1 sequence and areas of hper-intensity with possible fluid-fluid levels in weighted T2, this characteristic is peculiar and highly indicative of ABC of the bone[8].
A differential diagnosis requires combining and correlating clinical,radiological and histiological aspects, while a biopsy is indispensible in order to arrive at a certain diagnosis.
ABC-solid has always been a topic of great interest in pathologies as it can be mistaken for malign tumors, principally giant cell or osteosarcoma due to the cellularity and the variable mitotic activity[9].
The histological characteristics found in the solid type, as described by Sanerkin, are the presence of a rich fibroblastic and fibrohistiocitria with giant similarly appearing osteoclastic cells together with fused cells. Giant cells differentiate among each other for their shape, dimension and variable number of nuclei(5 to 30). This apparent histological tranquility does not agree well with the noteworthy local aggression of the lesion from both a radiological and clinical points of view. In fact, in several cases it mimics a sarcomatosis lesion[10].
The differing macroscopic and histiological aspects of ABC-solid and the classic variant include that the former of the two has a cystic cavity absent of blood but full of citrin liquid and a presence of an adherent membrane on the wall with an infiltrating appearance

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In fact, ABC-solid has a hemorrhagic cavity covered by a reddish-brown-membrane which can be easily removed and scarcely adhesive to the walls,he cortical bone. with no tendency to infiltrate
This different macroscopic aspect of ABC-solid is due to the presence of a tissue at histology varying from that observed in the classic variant. While ABC-classic is full of vascular lacune whose rapid formation swell the cortical to the point of provoking its destruction and in turn an intense periostal reaction that produces a sclerotic border, the solid variant is made up of a more solid tissue and richer in giant cells that evidence a slow growth, prevalently of an erosive nature[11].
A correct diagnosis is important for both selecting an appropriate surgical treatment and avoid disease recurrence. The simple removal of the cystic membrane in the solid variant is insufficient, though it is necessary to also remove the part of the tissue hiding in the niches of the cortical bone erosion
The natural course of ABC-solid type entails a growth and a continuous local destruction,despite this it can not be considered a pre-cancerous lesion, the recurrence rate of bone lesions is generally $\geq$ 20-25%.
The treatment of ABC depends on the location, vascularization, size and extension of the lesion as well as the patient's age. The inactive lesions can be monitored over time and can heal spontaneously. The active or aggressive lesions are at risk of recurrence and are thus treated with currettage and a treatment of the walls with adjuvants with or without grafting, whereas osteosynthesis is performed when bones are fragile[13]. The solid variant of ABC is more aggressive than the classic type, and for this a complete excision is necessary
Resectioning in block is indicated for eccentric lesions or in the case of lesions that involve bones that can be sacrificed. The resectioning in block and the substitution of removed bones with tricortical grafted transplants is a safe procedure with a minimal risk of recurrence and can be considered the treatment of choice if the resection is possible and the lesion less than 6-7cm [14]. Other authors affirm that the use of cement is an alternative to bone transplanting he so to reduce the of recurrence, minimize thermal necrosis and more easily refill the hole created due to the removal of the cyst[15]. According to several authors, generally after two years it is advised to fill the cavity by substituting the cement with grafted autoplastics once the recurrence has been excluded. In our case the lesion underwent curettage and was subsequently filled with cement, We have not substituted the cement with grafted autoplastics due to the optimal health of the patient.
Regarding the higher recurrence rate associated with ABC-solid compared to the classic variant, curettage needs to be extremely accurate and always accompanied by the use of local adjuvants: unlike cysts localized in long bones where the simple curettage is considered to be sufficient [16].
Further alternatives have been suggested and include adjuvant cryo-surgery followed by curettage and the curettage associated with an intra-lesional treatment of high speed milling or instillation of liquid nitrogen followed by a bone transplant.
The site of the lesion conditions the type of treatment. In fact it becomes more complicated if the lesion is localized in long bones, includes the growth cartilage, in several of the pelvic zones and vertebral column. Radiotherapy is not indicated except for cases insufficient rubbing, the embolization can be utilized in the vertebral locations and in the lesions particularly vascularized in association with curettage. The utilisation of demineralised bone matrix and the direct intra-lesional injection of adjuvant substances as primary therapy seems promising [17].
To reduce to a minimum the surgical intervention, a precise and immediate diagnosis of the lytic bone lesions with a sclerotic border and edema of the surrounding soft tissues is important.

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HUMAN AND ANIMAL RIGHT: For this type of study is not required any statement relating to studies on humans and animals.

References

1. Sanerkin NG, Mott MG, Roylance. Cancer. An unusual intraosseous lesion with fibroblastic, osteoclastic, osteoblastic, aneurysmal and fibromyxoid elements."Solid" variant of aneurysmal bone cyst. 1983; 51:2278-2286.

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2. Mascard E, Gomez-Brouchet A,Lambot K. Bone cysts: unicameral and aneurysmal bone cyst. Orthop Traumatol. Surg Res.2015 Feb ; 101(1 Suppl):S119-27. doi: 10.1016
3. Kransdorf MJ, Sweet DE. Aneurysmal bone cyst: concept, controversy, clinical presentation, and imaging. AJR Am J Roentgenol. 1995 Mar;164(3):573-80.
4. Bertoni F, Bacchini P, Capanna R, Ruggieri P, Biagini R, Ferruzzi A, Bettelli G, Picci P, Campanacci M. Solid variant of aneurysmal bone cyst. Cancer. 1993 Feb 1;71(3):729-34.
5. Parashari UC, Khanduri S, Upadhyay D, Bhadury S, Singhal S. Radiologic and pathologic correlation of aneurysmal bone cysts at unusual sites. J Cancer Res Ther. 2012;8(1):103-5.
6. Aycan OE, Çamurcu İY, Özer D, Arıkan Y, Kabukçuoğlu YS. Unusual localizations of unicameral bone cysts and aneurysmal bone cysts: A retrospective review of 451 cases. J Cancer Res Ther. 2012;8(1):103-5.
7. Glazebrook KN, Keeney GL, Rock MG. Ultrasound of primary aneurysmal bone cyst. Case Rep Radiol. 2014;2014:101069.
8. Mahnken AH, Nolte-Ernsting CC, Wildberger JE, Heussen N, Adam G, Wirtz DC, Piroth W, Bücker A, Biesterfeld S, Haage P, Günther RW. Aneurysmal bone cyst: value of MR imaging and conventional radiography. Eur Radiol. 2003;13(5):1118-24.
9. Lekka JA, Gavresea TV, Stanc-Giannakopoulos GA, Demertzis NS. Solid variant of aneurysmal bone cyst of the heel: a case report. J Med Case Rep. 2011 12;5:145.
10. Mendenhall WM, Zlotecki RA, Gibbs CP, Reith JD, Scarborough MT, Mendenhall NP. Aneurysmal bone cyst. Am J Clin Oncol. 2006 Jun;29(3):311-5.
11. Doğanavşargil B, Ayhan E, Argin M, Pehlivanoğlu B, Keçeci B, Sezak M, Başdemir G, Öztop F. Cystic bone lesions: histopathological spectrum and diagnostic challenges. Turk Patoloji Derg. 2015;31(2):95-103.
12. Cottalorda J, Bourelle S. Modern concepts of primary aneurysmal bone cyst. Arch Orthop Trauma Surg. 2007 Feb;127(2):105-14.
13. Tomasik P, Spindel J, Miszczyk L, Chrobok A, Koczy B, Widuchowski J, Mrozek T, Matysiakiewicz J, Pilecki B. Treatment and differential diagnosis of aneurysmal bone cyst based on our own experience. Ortop Traumatol Rehabil. 2009 Sep-Oct;11(5):467-75.
14. Iltar S, Alemdaroğlu KB, Karalezli N, Irgit K, Caydere M, Aydoğan NH. A case of an aneurysmal bone cyst of a metatarsal: review of the differential diagnosis and treatment options. J Foot Ankle Surg. 2009 Jan-Feb;48(1):74-9.
15. Lindfors NC, Heikkilä JT, Koski I. Bioactive glass and autogenous bone as bone graft substitutes in benign bone tumors. J Biomed Mater Res B Appl Biomater. 2009 Jul;90(1):131-6.
16. Rumi Takechi, Takashi Yanagawa, Tetsuya Shinozaki. Solid variant of aneurysmal bone cyst in the tibia treated with simple curettage without bone graft: a case report .World Journal of Surgical Oncology. 2012;10:45
17. de Gauzy JS1, Abid A, Accadbled F. J Pediatr Orthop B. Percutaneous Ethibloc injection in the treatment of primary aneurysmal bone cysts. 2005;14(5):367-70.

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