Acta Biomed 2018; Vol. 89, N. 2: 269-273 DOI: 10.23750/abm.v89i2.5278

Case report

Periosteal osteoblastoma of the distal fibula with atypical radiological features: a case report

Alessio Biazzo[1], *Elisabetta Armiraglio*[2], *Antonina Parafioriti*[2], *Norberto Confalonieri*[1]
[1] Orthopaedic Department, Gaetano Pini-CTO, Milano; [2] Pathology Department, Gaetano Pini-CTO, Milano

Summary. We reported the case of a 22 year-old boy who suffered a periosteal osteoblastoma of the distal fibula. The radiographic features of our case did not correlate with the majority of periosteal osteoblastomas of the long bones reported in the literature and were identical to a periosteal aneurysmal bone cyst. Periosteal osteoblastoma is a very rare tumor with a wide range of clinical and radiological features, showing in 15% of cases association with secondary aneurysmal bone cyst. Radiologist and orthopaedic surgeon should be aware of the atypical behavior of this rare entity in order to avoid mistakes with other more common tumors arising on the surface of the long bones. (www.actabiomedica.it)

Key words: periosteal osteoblastoma, aneurysmal bone cyst, atipycal, bone tumors

Introduction

Periosteal osteoblastoma (OBL) is a very rare tumor and was first reported by Lichtenstein and Sawyer in 1964 (1); then only few cases have been described in the english literature (2-10). OBL can be associated with aneurysmal bone cyst (ABC), which in this case represents a secondary lesion. The first description of this possibly association was attributed to Jaffe who described the possibility that an ABC might sometimes represent a secondary "blowout" in a preexisting bone lesion (11,12). The most frequent associations are with giant cell tumor, chondroblastoma and OBL, but more rarely even with osteosarcoma (OS) and fibrous dysplasia. In a review of 55 cases of OBL, Della Rocca and Huvos reported the presence of a secondary ABC in 15% of patients (13).

We described a case of periosteal OBL with radiological findings which resemble an ABC. The radiographic features of our case did not correlate with the imaging features that have been described in the majority of periosteal OBL of the long bones and were identical to a periosteal ABC. Histological examination confirmed the presence of secondary ABC.

Case-report

We report the case of a 22 year-old boy who was admitted at our Institute in September 2015 because of a 3 month-history of pain and swelling in the right ankle without previous trauma. Family history was unremarkable. The patient was studied with X-rays, Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) with contrast medium. X-rays (figure 1) showed a round eccentric osteolytic lesion of the anterior aspect of the distal fibula, without periosteal reaction or calcifications and with the typical shell of a cystic lesion. The CT (figure 2) confirmed a round osteolytic lesion of the anterolateral aspect of the fibula, arising from the surface of the bone. MRI showed multiple fluid-fluid levels (figure 3). The symptoms,

The study was carried out at the Orthopaedic Department of Gaetano Pini-CTO Institute in Milan

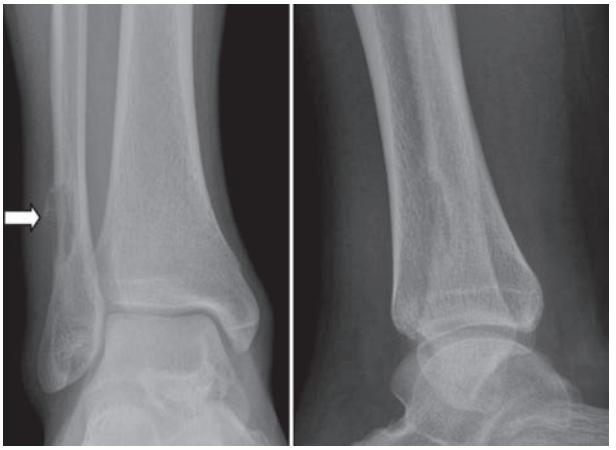

Figure 1. X-rays. On the left: antero-posterior view. Typical shell of cystic lesion (arrow). On the right: latero-lateral view showing an osteolysis of the anterior aspect of the fibula without bone sclerosis

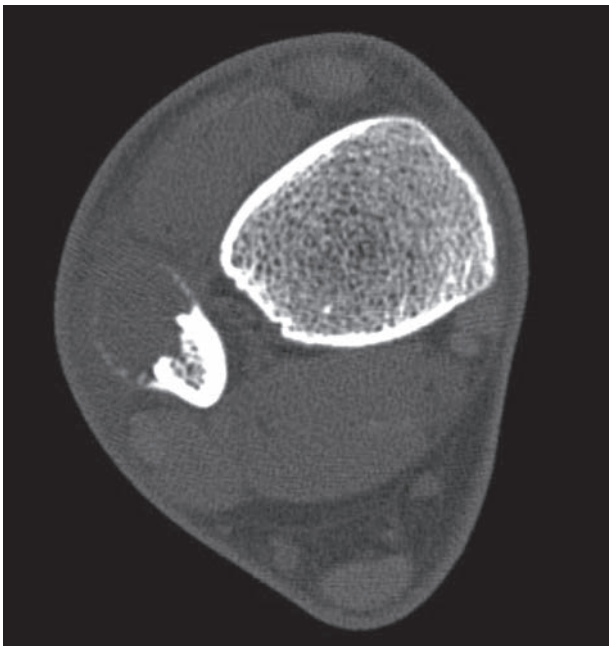

Figure 2. Non-contrast computed tomography showing an osteolytic lesion with thinning and infiltration of the cortex

age of the patient and imaging aspect were typical for ABC, so curettage was scheduled on October 2015. Intraoperative presentation was that of a solid and reddish lesion, without the aspect of an ABC; so an incisional biopsy was performed. The diagnosis was OBL with secondary ABC (figure 4). In November 2015 a curettage and packing with polymethilmethacrylate

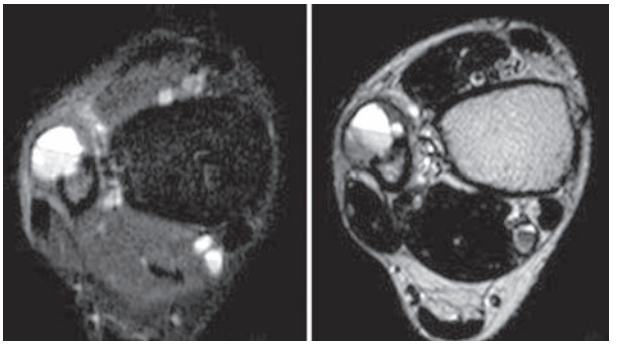

Figure 3. STIR-weighted (on the left) and T2-weighted (on the right) Magnetic Resonance Imaging after application of gadolinium showed fluid-fluid levels that are suggestive of aneurysmal bone cyst

was performed. We preferred polymethilmethacrylate because it offers a radiopaque interface on which a recurrence of disease can readily be determined. Postoperative x-ray did not show local recurrence (figure 5). The patient did not complaint pain or limitations.

Discussion

OBL is a benign tumor made of osteoblasts producing osteoid and woven bone.

It accounts for less than 1% of all bone tumors (14) and prefers males (2-3:1). Rarely observed before 10 and after 30 years of age. OBL shows evident predilection (50%) for the vertebral column (posterior arch) and the sacrum but it may occur in any skeletal site (15). In the long bones (1/3 of the cases) the proximal femur is the most frequent site of involvement (14,16,17). Swelling and pain are the clinical features of OBL. From a radiographic point of view it tends to be roundish, with margins often demarcated by a rind of bone sclerosis, not as dense as in osteoid osteoma. The cortex may be destroyed with intense periosteal reaction. The diagnosis relies on the histopathologic examination of the lesion (18). Most OBLs are active lesions, stage 2 according according Enneking classification (19). Occasionally, they are more invasive, bulging into the soft tissues (stage 3). Rarely the tumor appears almost quiescent and heavily mineralized, so that it can be approximated to a stage 1 lesion. In stage 1 (latent) or stage 2 (active), intralesional curettage with

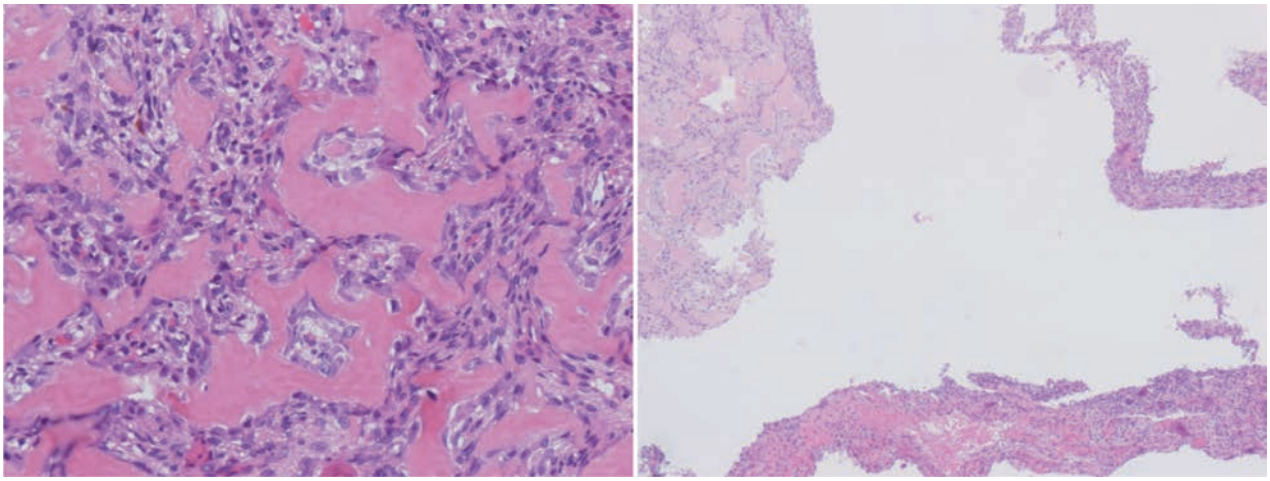

Figure 4. On the left: the tumor is characterized by a benign bone-forming proliferation with irregular osteoid deposition, haphazardly arranged spicules and trabeculae of woven bone lined by plump appositional osteoblasts. Vascularity is rich and osteoclast-like multinucleated giant cells are present. On the right: the tumor contains blood filled cystic spaces with fibrovascular septa, reactive bone formation and giant cells, indistinguishable from aneurysmal bone cyst (ABC). The diagnosis was osteoblastoma with ABC-like cystic changes

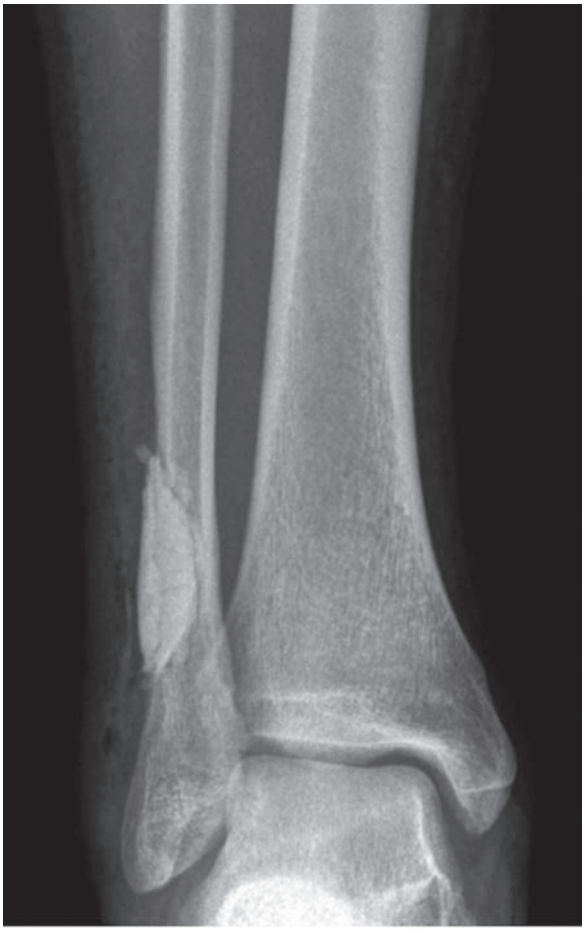

Figure 5. Post operative x-ray at 3 months

local adjuvants is used. In stage 3 lesions, (aggressive) marginal or wide resection is indicated (15).

Primary and secondary ABCs represent two different entities; in fact, secondary ABC show features that are more closely related to those of the associated lesion (20). Recent cytogenetic studies have shown clonal rearrangements of chromosomal bands 16q22 and 17p13, indicating a neoplastic basis in at least some primary ABCs (21-23).

The radiographic features of our case did not correlate with the imaging features that have been described in the majority of periosteal OBLs of the long bones. Most of the lesions presented with either a heavily mineralized mass, or a lytic lesion with central calcifications, on the surface of the diaphysis or metadiaphysis of a long bone. The lesions arose on the surface of the bone, with no evidence of cortical destruction, and were commonly associated with cortical thickening and benign periosteal new bone formation (24). Our OBL arose on the surface of the fibula, was a lytic, eccentric lesion, with an egg-shell calcification on the periphery, without periosteal reaction and blowing the cortex like an ABC. CT showed a lytic lesion thinning and infiltrating the cortex. No fluid-fluid levels could be appreciated with non-contrast CT. On contrast-medium MRI, the whole lesion was represented by fluid-fluid levels, which probably reflected intralesional haemor-

rages of different ages and are typical of ABC. This association was confirmed on histological slides.

The final diagnosis of OBL cannot be made based only on medical history, physical examination and imaging test findings. Pathological evaluation of the lesion should be included for a definitive diagnosis. Periosteal OBL needs to be differentiated from the following bone lesions arising on the surface of the long bones, such as parosteal and periosteal OS, high grade OS of the surface, ABC, periosteal chondroma and osteoid osteoma.

Parosteal OS is a slow-growing mass located mainly on the posterior aspect of the distal femur in adult patients (third-fourth decades of life). Clinical history is characterized by pain or swelling for months or years. On histological point of view, is characterized by spindle-cell stroma with minimal cytological atypia and rare mitosis, associated with long trabeculae of osteoid and woven bone (2).

Periosteal OS is a primary bone tumor of low-intermediate grade of malignancy affecting young patients (second decade of life) and arising mainly in the diaphysis of the long bones, with predilection for the proximal tibia and femur. It arises beneath the periosteum, elevating it and provoking new bone formation, which results in a radiolucent lesion on the bone surface with perpendicular striae. It is characterized by osteoid matrix and chondroid areas with anaplastic cells (2).

High grade OS of the surface is a high-grade malignant bone tumor arising on the bony surface, with minimal involvement of the underlying cortex. Apart from its site, does not differ from conventional intramedullary OS in age, histology and treatment. It shows predilection for the diaphysis of the femur.

ABC is a benign bone lesion usually found during the second decade of life. Most common sites are the metadiaphyses of the long bones and the spine. Few month-history of pain and swelling is the main clinical feature. From a radiographic point of view is an eccentric or subperiosteal, poor defined osteolysis, elevating and inflating the periosteum and progressively eroding the cortex. CT and MRI are often helpful in showing fluid-fluid levels within the cyst (15).

Periosteal chondromas usually develop during the second and third decades of life. They are located at the metaphysic of the long bones as well as in the hands and feet. Radiographs show a soft tissue mass with focal calcifications (2).

Osteoid osteoma has usually cortical or subperiosteal location in the long bones (femur and tibia) of young patients (15-25 years). The classical symptoms of this tumor, such as nocturnal pain which decreases with salycilates, are not present in OBL. Radiographs show bone sclerosis surrounding a central radiolucency which contains the "nidus", that is the active part of the lesion. Histologically, it resembles OBL, but the lesion does not exceed 2 centimeters.

Conclusions

Periosteal OBL is a very rare tumor with a wide range of clinical and radiographic features, showing in 15% of cases association with secondary ABC (13). A histological analysis is mandatory to get to a correct diagnosis, avoiding mistakes with other more common tumors arising on the surface of the long bones, such as parosteal and periosteal OS, high grade OS of the surface, ABC, periosteal chondroma and osteoid osteoma.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

References

1. Lichtenstein L, Sawyer WR. Benign osteoblastoma: further observations and report of twenty additional cases. J Bone Joint Surg Am 1964; 46: 755-65.
2. Sulzbacher I, Puig S, Trieb K, Lang S. Periosteal osteoblastoma: a case report and a review of the literature. Pathol Int 2000; 50(8): 667-71.
3. Patel S, Agrawal A, Maheshwari R, Chauhan VD. Periosteal osteoblastoma of the pelvis: a rare case. Iran J Med Sci 2015; 40(1): 77-80.
4. Kaya A, Altay T, Sezak M, Ozturk H. Periosteal osteoblastoma of the distal femur. Acta Orthop Belg 2009; 75(2): 280-5.
5. Mortazavi SM, Wenger D, Asadollahi S, Shariat Torbaghan S, Unni KK, Saberi S. Periosteal osteoblastoma: report of a

case with a rare histopathologic presentation and review of the literature. Skeletal Radiol 2007; 36(3): 259-64. Epub 2006 Jul 26.
6. Nakatani T, Yamamoto T, Akisue T, Marui T, Hitora T, Kawamoto T et al. Periosteal osteoblastoma of the distal femur. Skeletal Radiol 2004; 33(2): 107-11. Epub 2004 Jan 9.
7. Kawaguchi K, Oda Y, Miura H, Watanabe T, Tsuneyoshi M, Iwamoto Y. Periosteal osteoblastoma of the distal humerus. J Orthop Sci 1998; 3(6): 341-5.
8. Gentry JF, Schechter JJ, Mirra JM. Case report 574. Periosteal osteoblastoma of rib. Skeletal Radiol 1989; 18(7): 551-5.
9. Chatterji P, Purohit GN, Ramdeo Bikaner IN. Benign osteoblastoma of the maxilla (periosteal). J Laryngol Otol 1978; 92(4): 337-45.
10. Farman AG, Nortjé CJ, Grotepass F. Periosteal benign osteoblastoma of the mandible. Report of a case and review of the literature pertaining to benign osteoblastic neoplasms of the jaws. Br J Oral Surg 1976; 14(1): 12-22.
11. Jaffe HL. Aneurismal bone cyst. Bull Hosp Jt Dis 1950; 11(1): 3-13.
12. Jaffe HL. Discussion following paper by Donaldson. J Bone Joint Surg 1962; 44A: 40.
13. Della Rocca C, Huvos AG. Osteoblastoma: varied histological presentations with a benign clinical course. An analysis of 55 cases. Am J Surg Pathol 1996; 20(7): 841-50.
14. Unni KK. Benign osteoblastoma (giant osteoid osteoma). In: Unni KK, editor. Dahlin's bone tumors. General aspects and data on 11,087 cases, 5th edn. Philadelphia: Lippincott-Raven; 1996; 131-142.
15. Picci P, Manfrini M, Fabbri N, Gambarotti M, Vanel D. Atlas of Musculoskeletal Tumors and Tumorlike Lesions: The Rizzoli Case Archive. New York: Springer; 2014.
16. Lucas D, Unni KK, McLeod R, O'Connor MI, Sim FH. Osteoblastoma: clinicopathologic study of 306 cases. Hum Pathol 1994; 25(2): 117-34.
17. Papagelopoulos PJ, Galanis EC, Sim FH, Unni KK. Clinicopathologic features, diagnosis and treatment of osteoblastoma. Orthopedics 1999; 22(2): 244-7; quiz 248-9.
18. Schajowicz F, McGuire MH. Diagnostic difficulties in skeletal pathology. Clin Orthop Relat Res 1989; 240: 281-310.
19. Enneking WF. A system of staging musculoskeletal neoplasms. Clin Orthop Relat Res 1986; 204: 9-24.
20. Martinez V, Sissons HA. Aneurysmal bone cyst. A review of 123 cases including primary lesions and those secondary to other bonepathology. Cancer 1988; 61(11): 2291-304.
21. Panoutsakopoulos G, Pandis N, Kyriazoglou I, Gustafson P, Mertens F, Mandahl N. Recurrent t(16;17)(q22;p13) in aneurysmal bone cysts. Genes Chromosomes Cancer 1999; 26(3): 265-6.
22. Herens C, Thiry A, Dresse MF, Born J, Flagothier C, Vanstraelen G et al. Translocation (16;17)(q22;p13) is a recurrent anomaly of aneurysmal bone cysts. Cancer Genet Cytogenet 2001; 127(1): 83-4.
23. Oliveira AM, Hsi BL, Weremowicz S, Rosenberg AE, Dal Cin P, Joseph N, et al. USP6 (Tre2) fusion oncogenes in aneurysmal bone cyst. Cancer Res 2004; 64(6): 1920-3.
24. Kroon HM, Schurmans J. Osteoblastoma: clinical and radiologic findings in 98 new cases. Radiology 1990; 175(3): 783-90.

Received: 10 March 2016
Accepted: 25 May 2016
Correspondence:
Alessio Biazzo
via Bignami 1, Milano
Tel. 0039 3476234772
E-mail: ale.biazzo@yahoo.it