



Case report

Solid variant of aneurysmal bone cyst on the distal extremity of the radius in a child[☆]



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ABSTRACT

The solid variant of aneurysmal bone cysts (ABC) is considered rare. It occurs with greater frequency in pediatric patients and in the tibia, femur, pelvis and humerus. We present a case of a metaphyseal lytic lesion on the distal extremity of the radius in a child whose radiograph was requested after low-energy trauma. The hypothesis of a pathological bone fracture secondary to an aneurysmal bone cyst was suggested. After biopsy, the child underwent intralesional excision without bone grafting and the histopathological findings were compatible with the solid variant of aneurysmal bone cyst.

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Variante sólida do cisto ósseo aneurismático na extremidade distal do rádio em uma criança

RESUMO

A variante sólida do cisto ósseo aneurismático (COA) é considerada lesão rara, ocorre com maior frequência nos pacientes pediátricos e nos ossos da tíbia, fêmur, pelve e úmero. Apresentamos o caso de uma lesão lítica metafisária na extremidade distal do rádio de uma criança em que, ao exame radiográfico feito devido a um trauma de baixa energia, foi aventada a hipótese de fratura em um osso patológico secundária a um cisto ósseo aneurismático. Após a biópsia, a criança foi submetida a ressecção intralesional sem interposição de enxerto e o exame histopatológico foi condizente com a variante sólida do cisto ósseo aneurismático.

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Palavras-chave:

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Neoplasias ósseas

Fraturas do rádio

Criança

[☆] Study carried out at the Service of Orthopedics and Traumatology; Hospital de Clínicas; Universidade Federal do Triângulo Mineiro; Uberaba; MG; Brazil.

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Introduction

The aneurysmal bone cyst (ABC) is an expansile pseudotumor lesion of unknown etiology, usually found in the tibia, femur, pelvis and humerus.¹ The solid variant of the ABC was described in 1983 by Sanerkin et al.² due to histological predominance of solid material in the aneurysmal bone cyst. It is considered rare, accounting for 3.4–7.5% of all ABCs, occurring more commonly in pediatric patients.³ Pain is the most common symptom, followed by mild edema that can precede the definitive diagnosis in up to 12 months.³ Radiography and CT scan images disclose an expansile osteolytic lesion indistinguishable from the ABC.⁴

The solid variant of aneurysmal bone cyst is characterized by fibroblast proliferation without any cell or nuclear pleomorphism, giant cells similar to osteoclasts rich areas, aneurysmal sinusoids, differentiated osteoclasts with osteoid production and occasional foci of degenerated calcifying fibromyxoid tissue.²

The differential diagnoses include simple bone cyst, reparative giant cell granuloma, hyperparathyroidism brown tumor, giant cell tumor and malignant primary tumors such as chondrosarcoma, osteosarcoma and Ewing's sarcoma.⁵

In this report we present the case of a patient with the solid variant of aneurysmal bone cyst diagnosed after fracture of the distal extremity of the radius, secondary to low-energy trauma.

Clinical case

A two-year-old girl was brought to the emergency room with wrist pain for two days after falling on the ground, according to the family. Parents denied episodes of fever.

Physical examination showed pain on palpation of the distal right radius, edema, limitation in passive rotation and flexion–extension movements of the wrist due to pain, absence of joint stiffness and inflammatory signs.

Plain anteroposterior and lateral radiographs were taken (Fig. 1), and disclosed the presence of lytic metaphyseal lesion, respecting the limits of the distal radial physis, predominantly homogeneous, with cortical thinning associated with dorsal and volar cortical discontinuity of the distal extremity of the radius. After the initial evaluation, a CT scan was requested (Fig. 2) and demonstrated more clearly the characteristics of the lesion. Bone scintigraphy showed a monostotic lesion with focal increased uptake. Thereafter, it was suggested the hypothesis of distal radius fracture in a pathological bone, probably having an ABC as primary lesion, with differential diagnoses such as unicameral bone cyst and telangiectatic osteosarcoma.

The child underwent lesion biopsy that showed absence of neoplasia, but without definitive diagnosis. We opted for surgical treatment with intralesional excision (curettage) associated with adjuvant electrocauterization without interposition of bone grafts and/or bone cement. The harvested material was sent for histopathology and, after surgical wound closure, the child was immobilized with antebrachio palmar plaster cast kept for six weeks.



Fig. 1 – Anteroposterior (a) and lateral (b) radiographic views showing the lytic lesion in distal radial metaphysis.

Histopathology showed sparse multinucleated giant cells, intermingled with partially calcified trabecular immature bone; absence of necrosis, mitotic figures and aneurysmal spaces; and no evidence of simple bone cyst. The histopathological aspect was suspicious of solid aneurysmal bone cyst, despite the absence of aneurysmal vascular spaces as seen on the microscope images (Fig. 3).

After eight weeks new radiographs were taken (Fig. 4). On the fourth postoperative month, new radiographs showed reactive marginal sclerosis, distancing of the initial lesion from the distal radial physis and cortical thickening – modifications consistent with inactive lesion (Fig. 5).

Discussion

The solid variant of aneurysmal bone cyst and reparative giant cell granuloma were primarily described in craniofacial bones and small tubular bones of the hand and foot.² They are considered reactive and non-neoplastic lesions, although they can lead to misdiagnosis of giant cell tumor, hyperparathyroidism brown tumor and osteosarcoma (usually fibroblastic or low-grade variant).⁶

Clinically, the patient had pain on palpation of the distal radius and mild edema after low-energy trauma, which led us to the hypothesis of wrist contusion or even a possible fracture (subperiosteal or torus) of the distal radius – subtypes commonly found in this age group. The radiographic finding of an expansile metaphyseal lesion with cortical thinning of the distal radius that respected the physis with no periosteal reaction suggested a pathologic fracture, probably with an aneurysmal bone cyst as the primary lesion.

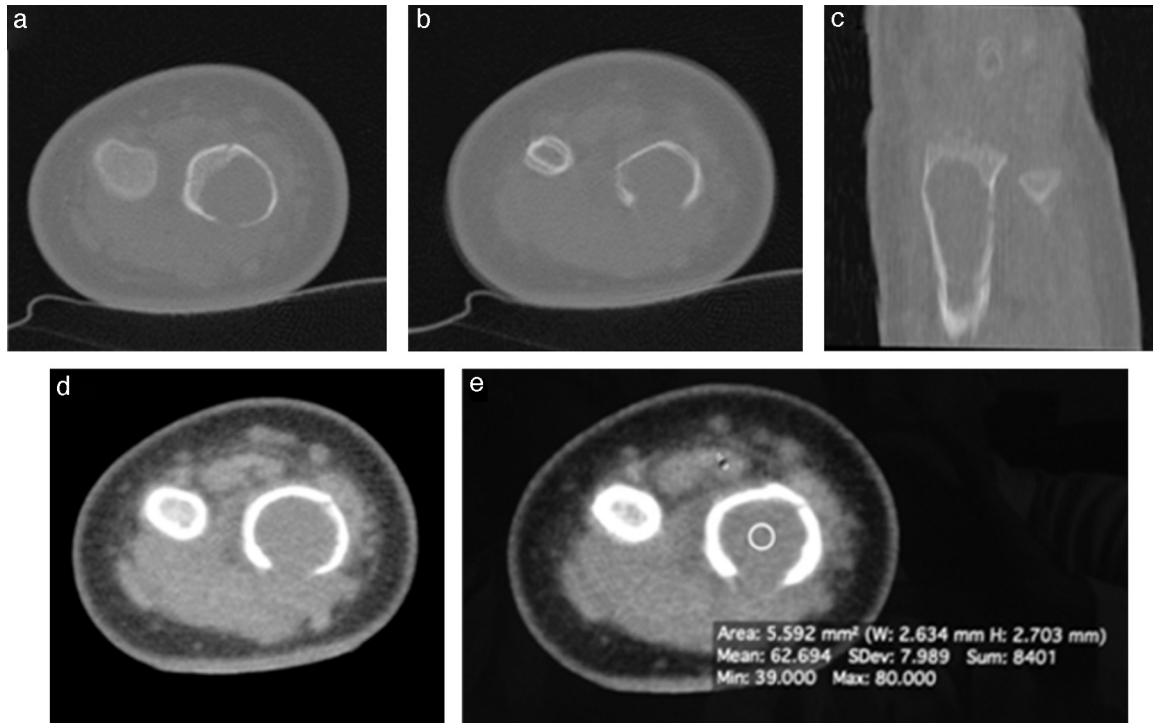


Fig. 2 – CT scan showing the expansile lytic lesion and associated fracture in the axial views (a and b); and the metaphyseal lesion length in the coronal view (c). In the axial view (d and e) with soft tissue window, attenuation of soft tissues within the intraosseous lesion that may represent a solid component or thick fluid content can be observed.

Although not specific, the detection of fluid level by CT scan and/or magnetic resonance imaging suggests the diagnosis of ABC.⁷ The presence of solid material inside the intraosseous lesion was identified by the authors by the analysis of the CT scan soft tissue window, although the diagnostic

confirmation of the solid variant of aneurysmal bone cyst was achieved only through the histopathological analysis, with the following pathologist description: ‘histopathological aspect is suspicious of solid aneurysmal bone cyst, despite the absence of aneurysmal vascular spaces. Immature cancellous bone

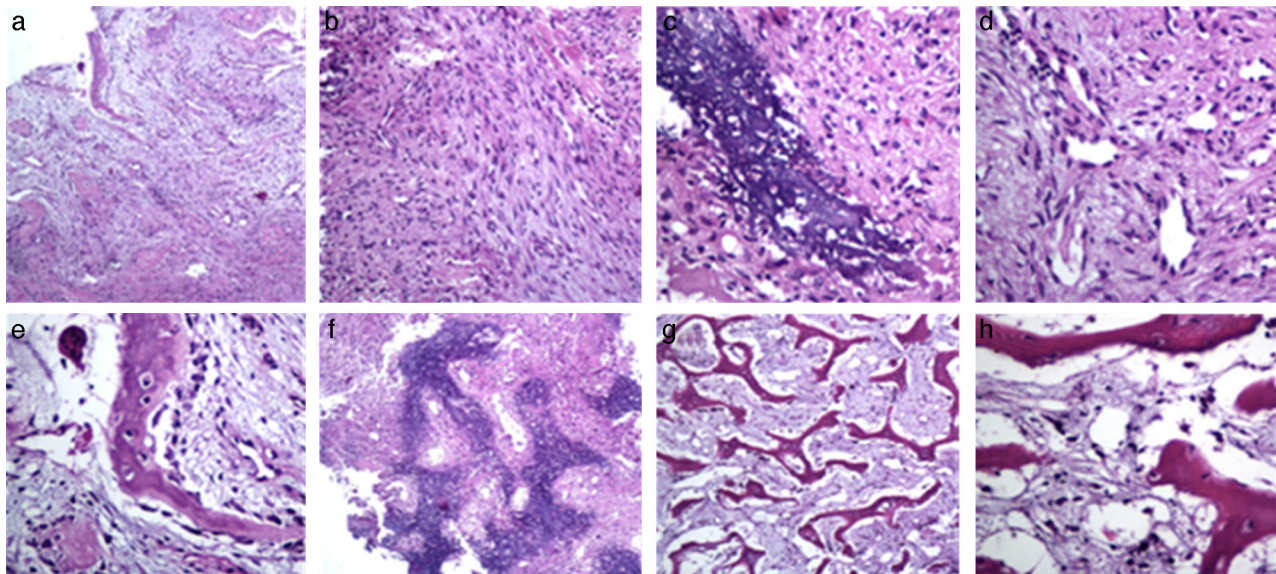


Fig. 3 – Histopathology images: moderately hypercellular, dense, fibrous connective tissue containing sparse multinucleated giant cells, with recent hemorrhage and intermingled with immature woven bone trabeculae partially calcified (a, b). Adjacent to the trabecular bone, abundant osteoblasts are often seen (c, d). Foci in the connective tissue where immature bone predominates (e). Region showing immature cancellous bone trabeculae with interstitial myxoid connective tissue (f-h).

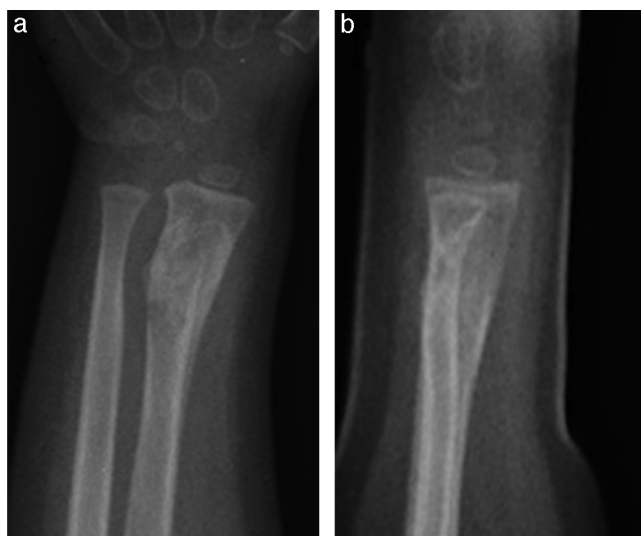


Fig. 4 – Anteroposterior (a) and lateral view (b) radiographs at six postoperative weeks showing fracture healing.



Fig. 5 – Anteroposterior (a) and lateral view (b) radiographs four postoperative months showing inactive lesion in the distal radial extremity.

with myxoid interstitial tissue is characteristic of bone fracture callus'.

Recent reports of the solid variant of aneurysmal bone cyst include the hamate bone as reported by Mavrogenis et al.,⁸ the

cervical spine by Karampalis et al.³ and the tibia by Takechi et al.⁹ Considering the scarcity of reports in the literature on the diagnosis of a solid variant of the ABC in the distal radius in a child, we considered relevant reporting this case.

Treatment recommendations for ABC include curettage associated with bone grafting. Considering the high recurrence rates these lesions have when treated with simple curettage only, many surgeons use adjuvant phenol or ethanol.

In the present case, considering the initial hypothesis of aneurysmal bone cyst and due to the proximity to the distal radial physis, we chose the intralesional curettage associated with adjuvant electrocauterization without graft interposition. Satisfactory evolution was observed during follow up, as shown by the present images with marginal sclerosis formation, distancing of the lesion from the distal radial physis and cortical thickening, consistent with lesion inactivity.

Conflicts of interest

The authors declare no conflicts of interest.

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