



Aneurysmal bone cyst-like areas as a sign of metastatic disease in the spinal column

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Aneurysmal bone cysts (ABCs) are vascular lesions with well-defined radiological signs. A fairly common MRI finding in both ABC and other primary bone tumors with high vascularity is the presence of cysts with fluid-fluid levels. Metastatic lesions with fluid-fluid leveling have not previously been defined in the literature. We report three cases of fluid-fluid leveling of the axial skeleton in metastatic lesions. Two patients had single-level and one multi-level neoplastic disease with distinctive cysts showing fluid-fluid levels on MRI evaluation. Diagnosis of gastric carcinoma in two patients and breast carcinoma in the third was achieved with trocar biopsies. At the time of this report all patients had completed initial oncologic treatments and were disease free.

Key words: Aneurysmal bone cyst; fluid-fluid leveling; spinal column; tumor.

Aneurysmal bone cysts (ABCs) are well-known vascular lesions of the bone consisting of blood-filled, anastomosing, cavernous spaces separated by cyst-like walls. It remains unclear whether their origin is dysplastic or neoplastic.^[1] Metaphyses of the long bones, as well as certain bones such as the calcaneus, are frequent locations for these lesions in the second and third decades of life. Involvement of more than one bone is unusual and there are no reports of multicentric occurrence in the literature.

Jaffe reported the possibility that ABCs might represent a secondary 'blowout' of some pre-existing bone lesion.^[2,3] Secondary ABC arising from mostly preexisting lesions of other aggressive benign tumors of the bone, such as giant cell tumors, osteblastomas, chondroblastomas, and fibrous dysplasia has been confirmed by other authors.^[1,4-6]

On standard radiographs, ABCs present as lytic lesions which expand the cortex of the host bone, usually leaving a thin but intact surrounding rim. Computed tomography (CT) scan is useful in defining the margin of the lesion and calcified bone shell, the pattern of osteolysis and the presence of fluid-fluid levels, which is a typical ABC feature, following contrast medium injection. Magnetic resonance (MR) imaging shows the fluid-fluid levels without contrast medium. The contents of the cystic cavities show a wide range of signal intensity on both T1- and T2-weighted images, likely reflecting intracystic hemorrhage of different ages.^[4,7] These findings, however, are not specific and reflect an underlying pathophysiology similar to that of ABC.

Aneurysmal bone cyst-like lesions in other conditions, such as metastatic lesion in the skeleton, are rare. To our knowledge, such a phenomenon in the spinal

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column has not yet been reported in the English speaking literature.

We report three patients with fluid-fluid leveling of the axial skeleton in metastatic lesions.

Case reports

Case 1

A 76-year-old female was admitted in April 2009 with a complaint of progressive back pain lasting for two months. The patient had no previous history of neoplastic disease. Standard radiograms were negative, although axial CT scan showed a Lodwick Grade 3 osteolytic lesion (permeated and moth-eaten) typical of a Stage 3 aggressive lesion (Fig. 1a). All of the multiple lesions were located in the anterior column as opposed to the more common posterior location of ABC. Frontal bi-dimensional electronic reconstructions revealed a pathological fracture of T2 at sectors 1 to 12 and layers A to D (WBB classification).^[8] MR imaging demonstrated multiple foci in vertebral bodies C6 to T2 and fluid-fluid levels in T2-weighted images. Bone scan and thoracic CT scan were negative for other lesions (Fig. 1b). The patient developed a sudden and rapidly progressive paraparesis immediately after presentation that necessitated emergency curettage and debulking with stabilization following selective arterial embolization through a transfemoral entrance and selective catheterization of the thyro-cervico-scapular trunks. A complete devascularization of the lesion could be achieved before surgery with the use of polyvinyl alcohol, facilitating surgical intervention. There were no other surgical complications and the final diagnosis was metastasis of breast adenocarcinoma with areas of ABC-like cysts. Six months following surgery, the patient remains under medical oncologic treatment and is asymptomatic with no neurological deficit (Frankel score E) and very good quality of life.

Case 2

A 67-year-old female was admitted in November 2009 with progressive back pain lasting for several months. The patient had a past history of gastric carcinoma ten years prior to admission and was under regular oncological follow-up with no recurrences or metastases. On standard AP radiographs, the right pedicle of L3 appeared to be lytic, suggesting destructive lesion although the lateral view was less specific with mostly preserved superior, inferior and anterior cortical layers. CT scans showed a Lodwick 1B lytic lesion with destruction of the right posterior arch along with a calcified shell highly suggestive of ABC. MR images showed a mass in the vertebral body of L3 at sectors 7 to 3 and layers A to D (WBB classification) and typical fluid-fluid levels with compression of the dural sac and roots (Fig. 2). A CT-guided biopsy performed was diagnostic for metastasis of gastric carcinoma with ABC-like areas. Thoracic CT scan showed multiple metastases with pleural effusion that required drainage. Bone scan showed multiple localizations as well. Final treatment was surgical curettage and stabilization after selective arterial embolization of the lesion. At the time of angiography, the lesion was vascularized by branches from the second and third lumbar arteries at the right. The patient was asymptomatic, without neurological deficit (Frankel score E) and well-satisfied at the 6th postoperative month. The patient remains under medical oncology treatment.

Case 3

A 53-year-old female presented in March 2010 with back pain for six months with no previous history of tumors. MRIs of the spine showed multiple lesions with fluid-fluid levels, located at the sacrum, L5 and T12 with a compression fracture at T12 (Fig. 3a). Chest and abdominal CT scan and bone scans were negative. The patient was initially diagnosed as a unique case of multi-

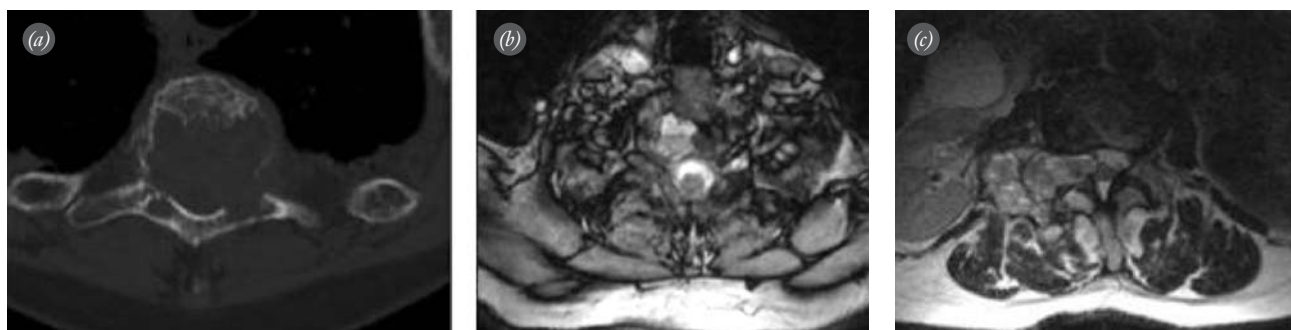


Fig. 1. Views from Case 1. **(a)** Axial CT scan demonstrating a lytic lesion extending to the foramen and destructing the anterior cortex of the lamina. **(b)** Axial T2-weighted MRI of the same lesion with typical fluid-fluid level in the cystic lesion.

Fig. 2. Axial T1-weighted image of the lesion from Case 2, demonstrating multiple fluid-fluid levels.

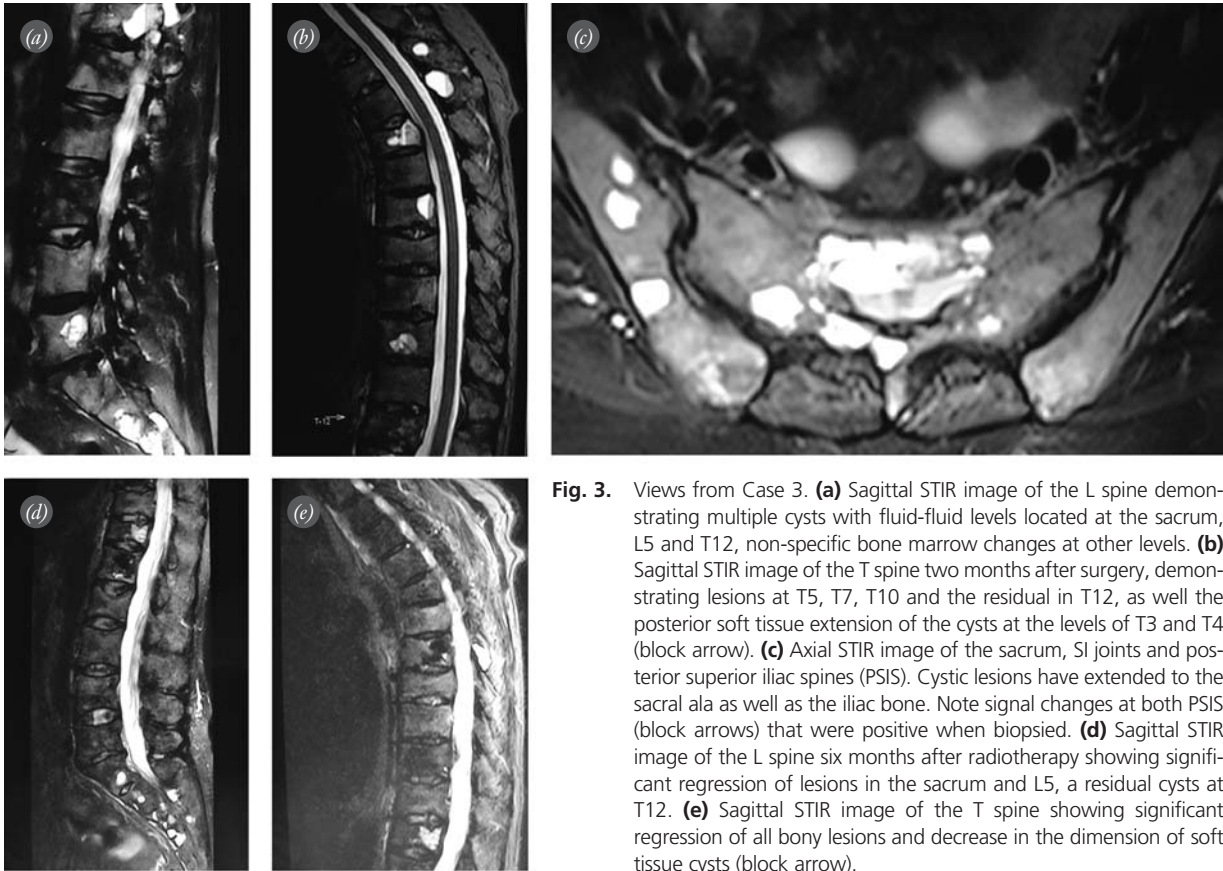


Fig. 3. Views from Case 3. **(a)** Sagittal STIR image of the L spine demonstrating multiple cysts with fluid-fluid levels located at the sacrum, L5 and T12, non-specific bone marrow changes at other levels. **(b)** Sagittal STIR image of the T spine two months after surgery, demonstrating lesions at T5, T7, T10 and the residual in T12, as well as the posterior soft tissue extension of the cysts at the levels of T3 and T4 (block arrow). **(c)** Axial STIR image of the sacrum, SI joints and posterior superior iliac spines (PSIS). Cystic lesions have extended to the sacral ala as well as the iliac bone. Note signal changes at both PSIS (block arrows) that were positive when biopsied. **(d)** Sagittal STIR image of the L spine six months after radiotherapy showing significant regression of lesions in the sacrum and L5, a residual cysts at T12. **(e)** Sagittal STIR image of the T spine showing significant regression of all bony lesions and decrease in the dimension of soft tissue cysts (block arrow).

level ABC of the spine with a compression fracture, which would explain her pain. Vertebroplasty of T12 and fluoroscopy-guided biopsies of T12 and sacrum were performed, revealing only nonspecific granulation tissue. In retrospect, it appears that these samples were obtained from within the cysts and cyst walls and not from the surrounding bony tissue. The patient was relatively well immediately following these surgical procedures but presented with increasing pain at two months. MRIs showed larger lesions at the L5 and sacrum, posterior element involvement at T3 and T4, new lesions at T7, T9, L1, L4 and bilateral posterior iliac crests, all with fluid-fluid levels (Figs. 3b and c).

The patient was restaged with a PET scan, revealing generalized increased uptake in the axial skeleton including bilateral iliac crests with SUV max values of 3.05 to 4 and a diffuse uptake at the right lobe of thyroid with a SUV max of 8.0. Due to the patient's level of pain, another series of vertebroplasties at the sacrum, L5, L4, L1, T11, T9 and T5 with simultaneous biopsies of the right iliac crest, L1, L4 and T9 were performed. Histology of the specimens taken from the iliac crests along with the pericystis tissue at L4 were positive for mucinous cell adenocarcinoma with ring cells, presum-

ably of gastric origin. Although endoscopy revealed only atrophic gastritis and failed to prove the stomach as the definitive primary location, the decision was made to treat the case as one of gastric carcinoma. The patient received palliative radiotherapy to her thoracic and lumbar spine and sacrum which decreased pain considerably. She received chemotherapy for 4 months. The patient's latest MRI at eight months revealed that most lesions resolved following radiotherapy (Figs. 3d and e).

Discussion

Aneurysmal bone cysts occurring in association with another condition are often assumed to represent a secondary change of a preexisting lesion. This view is largely based on clinical and radiologic observation of the sudden expansion of a previously relatively quiescent lesion. An alternative view is that the aneurysmal cystic element is an inherent part of the lesion, and that it has been present from the beginning.^[1] Our cases confirm that ABC can be present in association with other types of bone lesions, such as giant cell tumor, chondroblastoma, osteoblastoma, chondromyxoid fibroma and fibrous dysplasia, as benign lesions and osteosarcomas as malignant.

Most physicians do not regard ABC as a neoplasm and explain the characteristic spaces as unspecified 'hemodynamic disturbance' or 'arteriovenous fistulae'.^[9] Considering the three cases reported here, this assumption is presumably correct and ABC-like lesions may arise in any form of highly vascular neoplasm residing in the bone. However, it is unclear as to the reason that all cases were confined to the axial skeleton.

Two of our patients had no previous history of cancer and one was apparently disease-free for 10 years. As ABC-like lesions are often considered 'incidental' during the staging procedure of a known primary, the diagnostic confusion probably resulted in a delay in diagnosis and treatment of the widespread disease in at least one patient (Case 3). Therefore, these cases also illustrate the importance of biopsy in all lesions of the spinal column regardless of 'typical' appearance. It is also interesting to note that those 'typical' lesions appeared in relatively atypical locations in two of our three patients; one in the anterior column (rare), and the other at multiple levels (unique). To our knowledge, this is the first report of cystic lesions with fluid-fluid levels mimicking ABC in association with metastatic disease of the spine.

In conclusion, findings of ABC-like cystic lesions and fluid-fluid levels may also be associated with metastatic carcinomas of the axial skeleton. A thorough staging of the patients and vigorous sampling of the

lesions may be warranted in patients with rapidly progressive symptoms and atypical locations.

Conflicts of Interest: No conflicts declared.

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