



SYMPTOMS OF METASTATIC DISEASE OF THE SPINAL COLUMN: LESIONS APPEAR LIKE ANEURYSMAL BONE CYSTS

SPİNAL KOLONU TUTAN METASTATİK HASTALIK BULGUSU: ANEVİRİZMAL KEMİK KİSTİ BENZERİ LEZYONLAR

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SUMMARY:

Objectives: Aneurysmal bone cysts (ABC) are vascular lesions with well-defined radiological signs. A fairly common MRI finding is the presence of cysts with fluid-fluid levels. This finding is also known to be associated with other primary bone-forming tumors of the bone with high vascularity. However, metastatic lesions with fluid-fluid levels have never been defined in the literature.

Patients and methods: 14 patients presenting with pain and/or neurological loss at three separate spinal centers were included in this study. Twelve had a single fluid-fluid level, and the others had various levels, on MRI.

Results: Diagnosis was achieved with trocar biopsies in all patients, and aneurysmal bone cysts were established in 11 cases in accordance with radiological diagnosis (78.6%). Three of the patients (21.4%) had metastatic lesions. Two were diagnosed as gastric carcinomas and the third as breast carcinoma. At the time of this report, they had all completed their initial oncological treatment and were disease-free.

Conclusion: Cysts with fluid-fluid levels may not be a feature only of primary bone tumors of the spinal column. This case study demonstrates that a high level of suspicion and biopsies may be warranted for all patients for definitive diagnosis.

Key words: Aneurysmal bone cyst, metastasis, spine

Level of Evidence: Retrospective clinical study, Level III.

ÖZET:

Amaç: Anevrizmal kemik kistleri radyolojik özelliklerinin iyi tanımlandığı vasküler lezyonlardır ve Manyetik Rezonans Görüntüleme (MRG)'de sıvı-sıvı seviyelerinin olduğu kistler sıklıkla görülür. Bu bulgunun, damarlanması fazla primer kemik tümörlerinde de olduğu bilinmektedir. Ancak literatürde sıvı-sıvı seviyesi veren metastatik lezyonlar hiç tanımlanmamıştır. Bu çalışmada, metastatik lezyonlar olmalarına rağmen sıvı-sıvı seviyesi veren vaka serisini sunulması amaçlanmıştır.

Hastalar ve metod: Bu çalışmada üç farklı omurga cerrahisi merkezine, ağrı ve/veya nörolojik hasarla gelen 14 olgu bu seriye dâhil edilmiştir. Olguların on ikisinde tek seviyede, diğer vakada birçok seviyede MRG' de sıvı-sıvı seviyesi görüntüsü saptanmıştır.

Sonuç: Radyolojik olarak hastaların tamamında anevrizmal kemik kisti düşünülmüş, yapılan perkütan biopsiye 11 (% 78.6)'inde radyolojik tanı ile uyumlu olarak anevrizmal kemik kisti tanısı konulmuştur. Üç (% 21.4) olguda ise biopsiye metastatik lezyon saptanmıştır. Bunlardan iki tanesi mide kanseri, diğeri meme kanseri tanısı almıştır. Hastaların tümü temel onkolojik tedavilerini almış ve kür elde edilmiştir.

Tartışma: Sıvı-sıvı seviyesi veren kistler spinal kolonda sadece primer kemik tümörlerinde görülmezler. Sunulan bu vakalar kesin tanı için şüpheli yaklaşım ve biyopsinin tüm vakalarda uygulanması gerektiğini ortaya koymaktadır.

Anahtar Kelimeler: Anevrizmal kemik kisti, metastaz, omurga

Kanıt Düzeyi: Retrospektif klinik çalışma, Düzey III

INTRODUCTION:

Aneurysmal bone cysts (ABC) are well-defined vascular lesions of bone, but it is not certain whether their origins are dysplastic or neoplastic⁸. These lesions are in the shape of blood-filled cavernous spaces separated by walls, similar to a cyst. The metaphyses of long bones and the calcaneus are typical places for these lesions, and they are generally detected in the second or third decade.

Jaffe suggested that ABCs could emerge as a secondary problem over an underlying bone lesion⁵⁻⁶. Also, other authors have supported this suggestion that lesions that can be defined as secondary ABCs may occur in the presence of diseases such as giant cell tumors, osteoblastoma, chondroblastoma and fibrous dysplasia^{1,4,7-8}.

In standard roentgenography, these lesions are lytic lesions that destruct the cortex, but a thin line surrounding the limits remains robust. CT imaging is useful for the identification of the lesion borders, and a typical image for ABCs shows fluid-fluid levels after entry of the contrast substance. In MRIs, an image with fluid-fluid levels is seen with an ABC lesion without a contrast substance. The inside of the cystic cavity is observed at different intensities in T1 and T2 sequences. The reason for this is possibly because bleeding into the lesion occurs at different times^{2,7}.

As for metastatic bone lesions, an ABC-like appearance in other bone lesions is quite rare, and therefore this can cause mistakes. A similar phenomenon in the spinal column has not been yet reported in the literature. In this case study, MRIs showing fluid-fluid levels in the spine were examined concurrently in three different spinal

surgery centers. Three cases were diagnosed with metastatic lesions and were tracked.

MATERIALS AND METHODS:

In three spinal centers (two in Turkey and one in Italy), 14 patients with fluid-fluid levels by MRI, that were pre-diagnosed with aneurysmal bone cysts and had pain and neurological deficit, were included in the study. Nine patients were female and five were male. The ratio of male/female was 0.56. The mean age was 59.3 ± 11.4 (44–76). The distribution of lesions according to the spinal levels is given in Figure-1.

All patients had back and lower back pain, and four of them had neurological deficit.

A trocar biopsy was percutaneously performed for all patients under computerized tomography. The results of the biopsies and the radiological findings were evaluated.

RESULTS:

Aneurysmal bone cysts were established in eleven cases by percutaneous biopsy in accordance with the radiological diagnosis (78.6%). Three of the patients (21.4%) had metastatic lesions. Two were diagnosed as gastric carcinoma and the third as breast carcinoma. At the time of this report, all patients had completed their initial oncological treatment and were disease-free.

The first case in which a metastatic lesion was detected was a 76-year-old female who had had progressive back pain for two months previously, and came to the center in April 2009. The patient had no history of neoplastic disease.

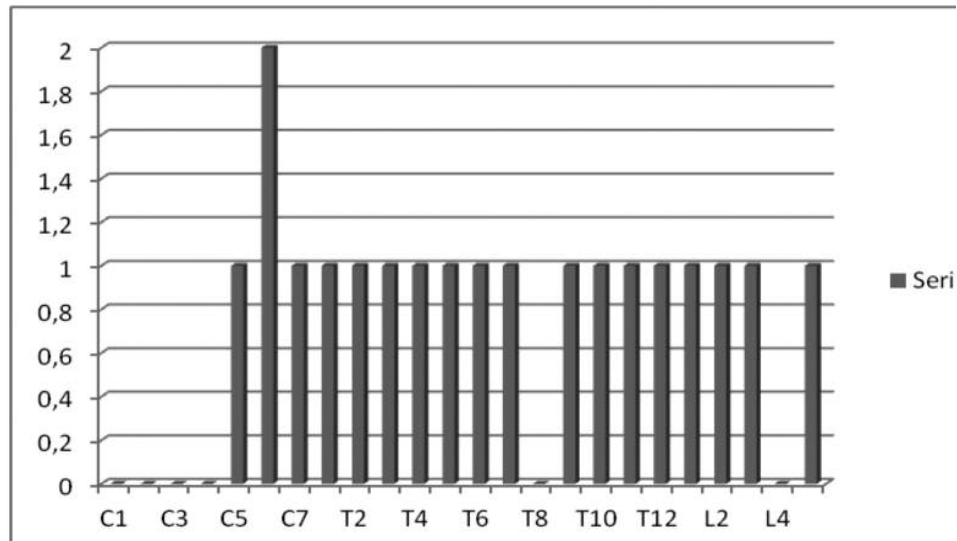


Figure-1. Distribution of lesions detected by MRI, according to the vertebra affected.

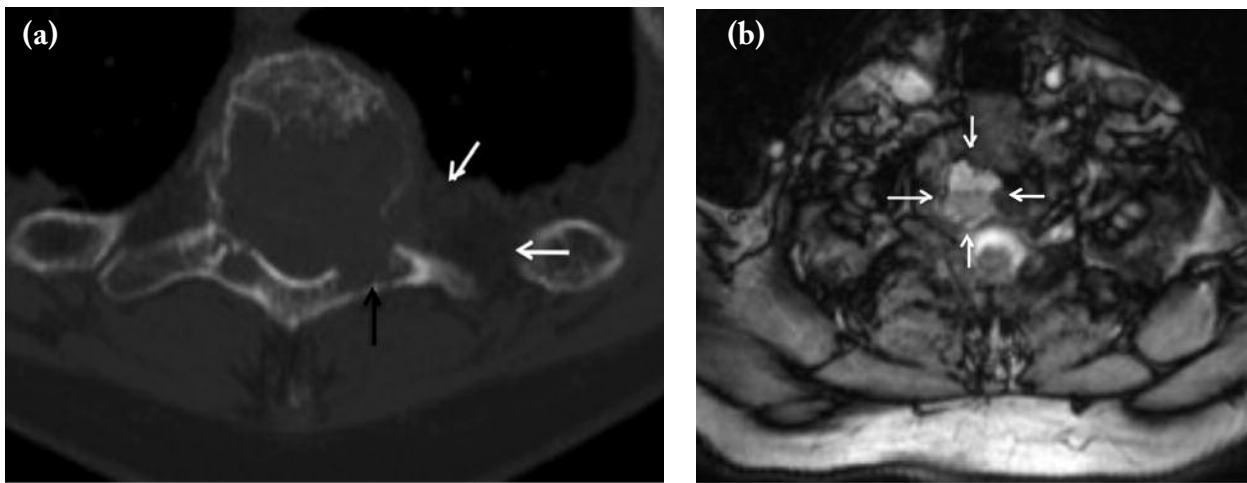


Figure-2. Case 1: **a)** Axial CT image showing a lytic lesion that extends to the foramen and destroys the front cortex of the lamina. **b)** Axial T2-weighted MR imaging showing fluid-fluid level in the same cystic lesion.

The patient had no pathological findings in standard roentgenography, but in axial tomography, Lodwick 3 osteolytic lesions (spread and moth-eaten view) were detected that were typical of stage 3 aggressive lesions (Figure-2.a).

Interestingly, all the multiple lesions were in the anterior column, differently to the posterior element involvement that is the

typical localization of ABCs. In frontal two-dimensional electronic reconstruction, a pathological fracture that extended from sectors 1 to 12 according to a WBB classification and held A, B, C, D layers was detected in the T2 vertebra³. In T2-weighted MR imaging, many centers with fluid-fluid levels were detected in the vertebral bodies from C6 to T2 (Figure-2.b).

No pathological findings were detected in bone screening or a thoracic CT in terms of other lesions. A rapid paraparesis developed, which started suddenly and immediately after the arrival of the patient to hospital. Therefore, stabilization with emergency curettage and a debulking procedure were trans-femorally applied to the patient after arterial embolization by tiro-cervico-scapular branch selective catheterization.

Complete avascularization of the lesion with polyvinyl alcohol before surgery made surgery easier. Other surgical complications were not encountered and the definitive diagnosis was chest adenocarcinoma metastasis, with cystic areas similar to ABC. The patient was still receiving oncological treatments six months later, had no complaints or neurological deficits (Frankel score E), and had a very good quality of life.

The second case in which a metastatic lesion was detected was a 67-year-old female who had progressive back pain for a few months previously and came to the center in November 2009. She had been diagnosed with gastric carcinoma ten years previously, had been followed up regularly, and there was no recurrence or metastasis until that time. In standard antero-posterior roentgenography, the L3 vertebra right pedicle had a lytic image considered to be a destructive lesion. A lateral X-ray contained non-specific findings that the upper, lower and frontal cortical structures were mostly protected. In CT imaging, there was a Ludwig 1B lytic lesion that showed right posterior arch destruction with a calcified shell, like an ABC. In MRI, there was a mass extending from sector 7 to 3 in the L3 vertebral corpus, holding A, B, C, D layers (WBB classification), giving a typical

fluid-fluid level and pressing on the dural sac and roots (Figure-3).

The diagnosis after biopsy with CT was metastasis of gastric carcinoma containing areas similar to ABCs. In thoracic CT screening, there were multiple metastases with pleural effusions that required drainage. Similarly, many centers were detected by bone screening. The treatment was surgical curettage and stabilization after selective arterial embolization of the lesion. During angiography, it was observed that the lesion took branches from the third and fourth lumbar arteries from the right. There were no problems in the postoperative period.

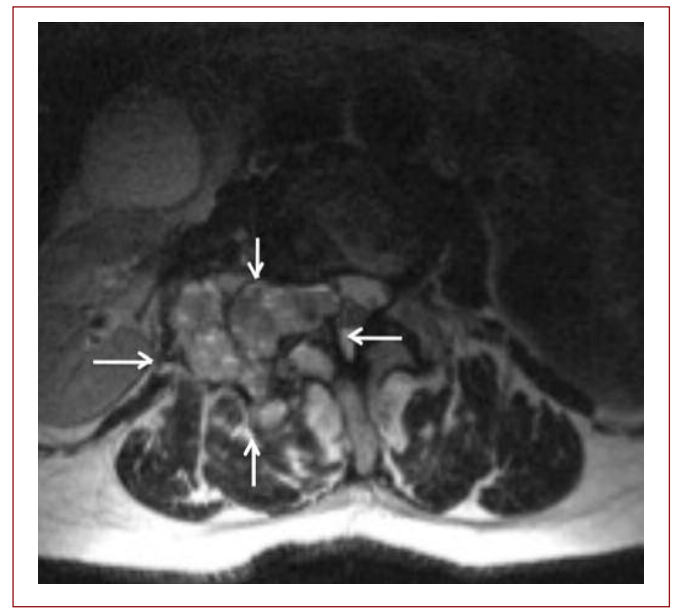


Figure-3. Axial T1-weighted MRI of case 2 showing lesions with many fluid-fluid levels.

At the end of six months, the patient was asymptomatic, there was no neurological deficit (Frankel score E), and the living standard was satisfactory. The patient still continues with oncological treatment.

The third case in which a metastatic lesion was detected was a 53-year-old female who had no

previous tumor history, had progressive back pain for six months previously, and came to the center in March 2009. In MRI, there were lesions with multiple fluid-fluid levels in the sacrum, L5, T5, T9 and T12, and there was also an image compatible with a compression fracture in T12 (Figure-4.a).

For staging, bone screening by chest and abdominal CT were performed, and there were no additional problems. As a result of these evaluations, the patient was considered to have multiple ABC lesions and a compression fracture. The pain was thought to be due to the compression fracture. Vertebroplasty at the T12 vertebra and a biopsy at T12 and the sacrum with fluoroscopy were performed. The pathological evaluation was compatible with nonspecific granulation tissue. On retrospective evaluation, it is thought that the biopsy samples were not from adjacent bone tissues, but from the inside of a cyst and the cyst wall. Although the patient's condition improved after surgery, she was re-admitted after two months with increased back pain. In an MRI taken at the second admittance, it was detected that lesions at the L5 and sacrum were enlarged, there was involvement of posterior elements at the T3 and T4 levels, and that new lesions were present at T7, T9, L1, L4, and both bilateral iliac wings, and they all had fluid-fluid levels (Figure-4.b,c).

Considering these results, a PET scan test was applied to stage the patient. Common up-take with a SUVmax value of 3.05-4 was detected in the axial skeletal and bilateral iliac wing, and another with a SUVmax value of 8.0 was observed in the right gland of the thyroid. A series of vertebroplasty was again performed to the sacrum, L5, L4, L1, T11, T9 and T5, due to the severe pain of the patient. In the

same session, biopsies were taken from the right iliac wing, L1, L4 and T9. The specimens taken from the iliac wing and the L4 pericyclic tissue were compatible with ring cell mucinous adenocarcinoma, which was thought to be of gastric origin. Although an endoscopic evaluation was compatible with only atrophic gastric, and did not support the idea that the primary tumor originated from the stomach, treatment planning of the patient was carried out by considering gastric carcinoma. Palliative radiotherapy was applied to the thoracic and lumbar vertebrae and sacrum, and the patient's pain was significantly reduced. After observation of any progression in the patient, chemotherapy was given for four months. In an MRI after eight months, it was observed that many lesions had regressed after radiotherapy and chemotherapy (Figure-4.d,e). At the last follow-up in May 2012, no change in the lesions was observed.

DISCUSSION:

In 14 cases with fluid-fluid level images in MRIs, metastatic lesions were detected in 21.4% of cases, unlike the radiological diagnoses. In these three cases, it was detected that metastatic disease of the spinal column was in the shape of cystic lesions with radiological and histological findings compatible with ABCs, giving fluid-fluid levels. This is the first description of metastatic lesions in the spine.

When an ABC is associated with another disease, many authors consider it to be a secondary change of the first lesion. This perspective is explained due to clinical and radiographic observations of sudden expansions of lesions that were previously silent.

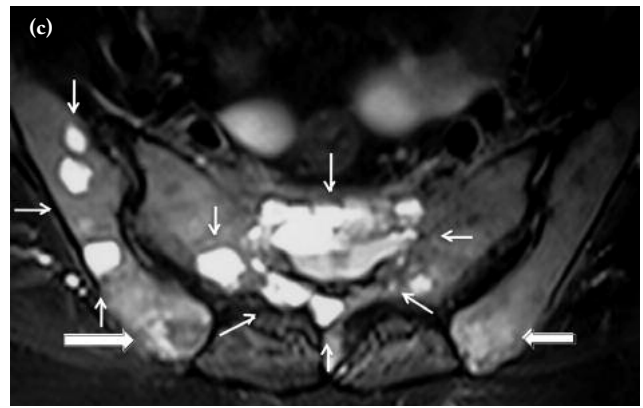
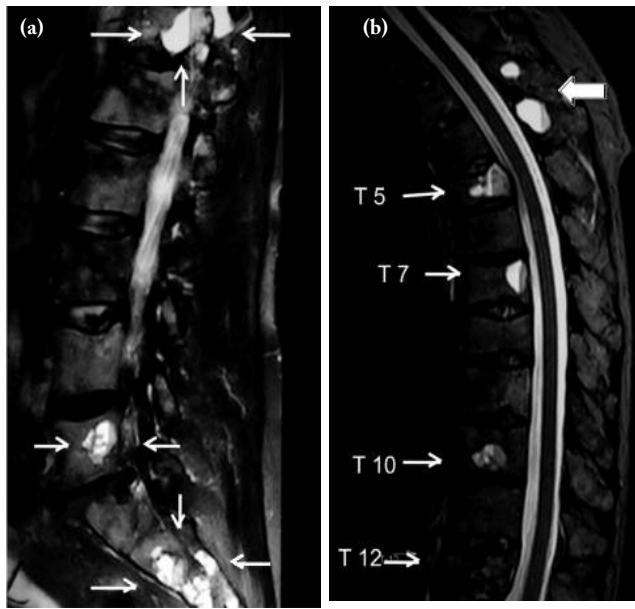


Figure-4. Case 3: **a)** Sagittal STIR images of lumbar vertebrae showing cysts with many fluid-fluid levels at the sacrum, L5 and T12 levels, and non-specific changes in the bone marrow at other levels. **b)** Sagittal STIR images of thoracic vertebrae, cystic lesions with fluid-fluid levels at the T5, T7, T10 and T12 vertebral corpus and posterior bone elements and soft tissue at the T3 and T4 levels. **c)** Axial STIR image of sacrum showing that the lesions distribute to the sacral ala and iliac bone. Note the signal change in the posterior superior iliac projection. Biopsy samples taken from this lesion were found to be compatible with metastasis in pathological evaluations. **d)** Six months after radiotherapy, sagittal STIR images of the lumbar spine show clear regression of the lesions at the sacrum, L5 and T12 levels. **e)** Sagittal STIR image of the thoracic spine showing clear regression of all bone lesions after radiotherapy. The soft tissue cysts seem to have become smaller in size.

Another alternative perspective is that aneurismal cyst elements are natural parts of the lesion and have existed from the beginning⁸.

According to our experiences, ABCs rarely occur in combination with certain bone lesions, including benign lesions such as giant cell tumors, chondroblastoma, osteoblastoma, chondromyxoid fibroma, fibrous dysplasia, and malign lesions such as osteosarcoma. Many

clinicians do not consider ABCs to be neoplasms, and characterize these gaps as “hemodynamic disorders” or “arteriovenous fistula”⁵.

In the light of the cases presented, this assumption is probably correct, and ABC-like lesions can occur in all neoplasms with increased vascularity that are established in bone. The explanation of lesions in all cases limited to the axial skeleton is still unknown.

In these three patients with metastases, it is remarkable that two had no history of cancer, and the other had had no occurrence of disease symptoms for ten years. This situation is contrary to diagnosis of a random lesion detected during a primary disease staging, which led to confusion and may have led to diagnosis and treatment delay, at least for one patient (the third case). These cases show that biopsies should be taken from all lesions, no matter how many typical lesions are present in the spinal column. Interestingly, typical lesions emerged in relatively atypical localizations in two of our cases.

A rarely-seen lesion located in the anterior column was detected in one case, while lesions detected at many levels have not been reported until now.

As a result, our three cases with ABC-like cystic lesions and fluid-fluid levels showed that these findings may be related to metastatic carcinoma of the axial skeleton. Therefore, metastatic malignant tumors should be considered in patients with ABC-like cystic lesions and fluid-fluid levels, and so screening of patients should be done carefully for atypically-located lesions with rapid progressive symptoms, and a biopsy should be taken from the lesion.

REFERENCES:

1. Balcı P, Obuz F, Göre O, Yılmaz E, Demirpolat L, Aktuğ T, Kovanlıkaya I. Aneurysmal bone cyst secondary to infantile cartilaginous hamartoma of rib. *Pediatr Radiol* 1997; 27(9): 767–769.
2. Beltran J, Simon DC, Levy M, Herman L, Weis L, Mueller CF. Aneurysmal bone cysts: MR imaging at 1.5 T. *Radiology* 1986; 158(3): 689–690.
3. Boriani S, Weinstein JN, Biagini R. Primary bone tumors of the spine. Terminology and surgical staging. *Spine* 1997; 22(9): 1036–1044.
4. Boriani S, De Iure F, Campanacci L, Gasbarrini A, Bandiera S, Biagini R, Bertoni F, Picci P. Aneurysmal bone cyst of the mobile spine: report on 41 cases. *Spine* 2001; 26(1): 27–35.
5. Jaffe HL. Aneurysmal bone cyst. *Bull Hosp Jt Dis* 1950; 11(1): 3–13.
6. Jaffe HL. Discussion following paper by Donaldson. *J Bone Joint Surg* 1962; 44A: 40.
7. Kransdorf MJ, Sweet DE. Aneurysmal bone cyst: concept, controversy, clinical presentation and imaging. *Am J Roent* 1995; 164(3): 573–580.
8. Martinez V, Sissons HA. Aneurysmal Bone Cyst; a review of 123 cases including primary lesions and those secondary to other bone pathology. *Cancer* 1988; 61(11): 2291–2304.

