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CASE REPORT

Bone metastases with multiple fluid-fluid levels from gastric cancer: a case report and review of literature

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Abstract

We report the case of a 54-year-old man with 6 months of progressive sacral pain. Computed tomography showed a large osteolytic mass in the sacrum with multiple osteolytic foci in the ilium and lumbar spine. Magnetic resonance imaging revealed multiple fluid-fluid levels in the sacral lesion as well as in the lumbar foci. The multiple bone lesions were initially diagnosed as a primary sacral malignancy with multiple bone metastases. A biopsy of the sacral lesion under the guidance of computed tomography was performed and a metastatic adenocarcinoma from the stomach or lung was found pathologically. Finally, a gastric tubular adenocarcinoma was detected by endoscopic biopsy and the diagnosis of the patient was gastric cancer with uncommon multiple bone metastases.

Keywords Metastasis · Gastric cancer · Fluid-fluid levels · Bone tumors

Introduction

Fluid-fluid level (FFL) is an important radiologic sign in the diagnosis of musculoskeletal tumors. It is most commonly seen and initially reported in aneurysmal bone cysts [1]. FFLs are non-specific and have been found in a variety of benign and malignant bone tumors, including giant cell tumors, fibrous dysplasia, Langerhans' cell histiocytosis, osteosarcoma, etc. [2]. However, FFLs are infrequently seen in skeletal metastases and have only been presented in 11 case reports since its first description by Collet et al. [3] in 1988. Here we report an uncommon case of bone metastases with multiple FFLs from a previously unknown gastric carcinoma, of which the sacral lesion mimics a primary bone tumor and was misdiagnosed as a sacral osteosarcoma with multiple bone metastases before biopsy.

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Case report

A 54-year-old man presented with progressive sacral pain lasting for 6 months. He had no previous neoplasm history. Laboratory abnormalities were as follows: alkaline phosphatase elevated to 764 U/l (normal < 125 U/l), osteocalcin and 25-OH-VitD_3 slightly decreased to 12.0 ng/ml (normal 14–46 ng/ml) and 9.0 ng/ml (normal ≥ 20 ng/ml) respectively, and fecal occult blood test was weak positive. Normal values were found for parathyroid hormone and serum tumor markers. Computed tomography (CT) scan showed an osteolytic lesion in the sacrum with a large soft tissue mass (Fig. 1a). High-density septations were found in the sacral lesion, while no osteosclerotic margins were seen (Fig. 1b). Multiple osteolytic foci were also found in the ilium, lumbar vertebrae, and posterior elements. Magnetic resonance imaging (MRI) revealed multiple FFLs inside the sacral lesion (Fig. 1c, d, e). The upper layers of the FFLs were hyper to iso intensity on T1-weighted images (T1WI), while the lower layers were iso to hypo intensity on T2-weighted images (T2WI). More FFLs were demonstrated on T2WI than on T1WI. FFLs were also seen in some of the foci in the lumbar spine. The separations and the peripheral solid part of the sacral lesion displayed an obvious enhancement after contrast injection on MRI (Fig. 1f). Almost the whole sacrum was involved, as well as the adjacent piriformis muscles.

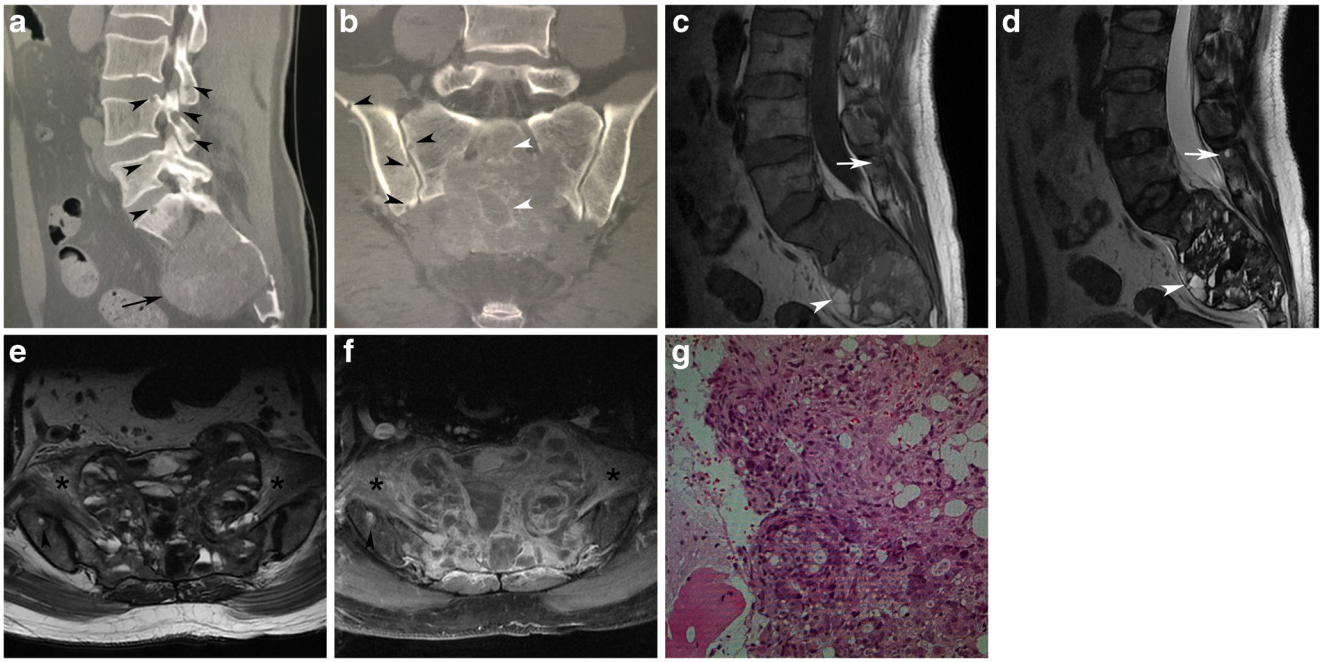

Fig. 1 **a** A 54-year-old man presented with 6 months of progressive sacral pain. Sagittal computed tomography image shows an osteolytic lesion with a large soft tissue mass (*black arrow*) in the sacrum. Multiple osteolytic foci are found in the lumbar vertebrae and posterior elements (*black arrowheads*). **b** High-density septations are revealed on coronal CT images (*white arrowheads*). Small osteolytic foci are also seen in the ilium (*black arrowheads*). **c** Several FFLs are revealed on the sagittal T1-weighted image (*white arrowhead*). Multiple lumbar foci (*white arrow*) are also depicted, as seen on CT images. **d** Sagittal T2-weighted image shows multiple FFLs inside the sacral lesion (*white arrowhead*). FFL is also seen in the lumbar foci (*white arrow*). **e** Axial T2-weighted image shows high signals in the adjacent muscles to the sacral mass (*black stars*). **f** Axial contrast-enhanced T1-weighted image with fat suppression reveals an obvious enhancement of the solid part of the sacral lesion as well as the adjacent muscles (*stars*) and the focus in the right ilium (*black arrowheads*). **g** Microscopy of the sacral biopsy specimen (H&E × 200)

A primary sacral bone malignancy (osteosarcoma or chordoma) with secondary aneurysmal bone cysts was initially considered and multiple bone metastases from the sacral tumor were diagnosed for the other lesions. A biopsy of the sacral lesion under the guidance of CT was made. A metastatic adenocarcinoma from the stomach or lung was pathologically diagnosed (Fig. 1g). The patient underwent following examinations of the chest and abdomen. Gastric tubular adenocarcinoma was affirmed by endoscopic biopsy, and the final diagnosis of the patient was gastric cancer with multiple bone metastases. The patient received chemotherapy and radiotherapy.

Discussion

The FFLs occurred in 2.7 to 11.2% of focal bone lesions [1], while they can be found in less than 5% of bone metastases [4]. Only nine case reports (with 11 cases in total) of bone metastases with FFLs were documented in detail in the English literature. A summarization of the reported cases is exhibited in Table 1. Most of the reported cases had multiple lesions (8 of 11) and the patients were older than 45 years old, as well as the present case. These are in line with the characteristics of bone metastases in literature [13]. Almost all of the reported bone metastases with FFLs originated from breast, lung, or gastric carcinoma except two cases (one from neuroendocrine carcinoma and the other from carcinoma with unknown origin), indicating the FFLs in bone metastases may provide useful clues of the primary malignancy, especially when no previous history of neoplasm was presented. Although gastric cancer infrequently metastasizes to bones, which has been reported in only 0.9 to 10% of the gastric cancer patients in clinical practice [14], the presence of FFLs in bone metastases might indicate a gastric origin.

The FFLs usually stand for the hemorrhage in the tumor [1] and may be found in any form of highly vascular bone neoplasm [10]. In malignant bone lesions, FFLs arise from tumorous and hemorrhagic necrosis with layers of different densities [8]. As plenty of intracellular mucus was found in the tumor cell pathologically, the formation of multiple FFLs in the present case might be partly due to the mucus secretion of the gastric adenocarcinoma cells. The FFLs inside the tumor can be better depicted on MRI than on CT images [15]. T2WI is the most frequently used sequence for its high sensitivity as shown in the present case and literature [10–12], and the FFL usually displays as high signal intensity of the upper layer and low signal intensity of the lower layer on T2WI. However, the

Table 1 Characteristics of 12 cases of bone metastases with fluid-fluid levels

Reference	Age/gender	Symptoms/ previous history of neoplasm	Location	Number of lesions/number of FFLs in one lesion	Origin of metastases
Lum et al. 1990 (abstract only) [5]	Unknown	Unknown	Rib	Unknown	Primary carcinoma of the bronchus
Nguyen et al. 1996 [6]	75/F	Pain and swelling of the right knee/breast adenocarcinoma	Distal femur	Single/multiple	Breast adenocarcinoma
Kickuth et al. 2002 [7]	61/M	Low back pain/undifferentiated small cell neuroendocrine carcinoma of the urinary bladder	Vertebrae of L4-5	Multiple/multiple	Undifferentiated small cell neuroendocrine carcinoma of the urinary bladder
Bladt et al. 2004 [8]	74/F	Low back pain, cauda equine syndrome, and decreasing strength in both legs/none	Thoracic/lumbar/sacral vertebrae	Multiple/multiple	Poorly differentiated carcinoma with unknown origin
Laurent et al. 2010 [9]	72/F	Pain in the thoracic and lumbar spine and the left tibia/none	Spine/pelvis/femur	Multiple/multiple	Breast lobular adenocarcinoma
Mohamed et al. 2013 [1]	47/M	Back pain/none	Posterior elements of L1	Single/single	Poorly differentiated lung adenocarcinoma
Karadeniz et al. 2013 [10]	76/F	Progressive back pain/none	Vertebrae of C6 to T2	Multiple/multiple	Breast adenocarcinoma
	67/F	Progressive back pain/gastric cancer	Thoracolumbar spine/pelvis	Multiple/multiple	Gastric adenocarcinoma
	53/F	Back pain/none	Spine (lumbar/thoracic/sacrum)	Multiple/multiple	Gastric adenocarcinoma
Colangeli et al. 2014 [11]	55/F	Back pain/none	Posterior arch of T12	Single/multiple	Lung adenocarcinoma
Gundogdu et al. 2016 [12]	49/F	Severe lower back pain/none	Vertebrae of thoracolumbar/ sacral spine	Multiple/multiple	Breast pleomorphic lobular carcinoma
Min et al. 2018 (the present case)	54/M	Progressive sacral pain/none	Spine/pelvis	Multiple/multiple	Gastric adenocarcinoma

M male

F female

signal intensity of the FFLs may vary in different cases for the different ages of blood deposits inside the tumor [9].

The sacral lesion with multiple FFLs in the present case was different from a primary aneurysmal bone cyst for its apparently enhancing soft tissue masses, the involvement of the adjacent muscles and the coexistence of the multiple foci in the other segments of the spine. Also, a primary aneurysmal bone cyst, which is most commonly found in the first two decades of life [16], is uncommon in patients of this age. However, the large sacral osteolytic lesion looked extremely like a primitive malignant sacral tumor for its size and the internal multiple FFLs, and an imaging appearance like this case has rarely been reported in previous cases of bone metastases. The sacral lesion was hard to be confirmed as a metastatic tumor without a biopsy, although there were multiple lesions in the other bone sites.

The differential diagnosis includes sacral chordoma, sacral giant cell tumor, or sacral telangiectatic osteosarcoma with multiple bone metastases.

Chordoma is the commonest primary sacral tumor and accounts for 40% of all sacral tumors. It is most commonly seen in the fifth and sixth decades of life and has a predilection for men [17]. The tumor is usually characterized by a very high signal on T2WI and multiple FFLs can also occur inside the tumor [2] due to hemorrhage and necrosis. However, distant metastases are uncommon in chordomas [17]. Giant cell tumor is another commonly seen primary tumor in the sacrum [18]. Typically, it is an osteolytic lesion without sclerotic rims and calcifications on CT images and has low signal intensity on both T1- and T2-weighted images [17]. Secondary aneurysmal bone cysts with multiple FFLs are frequently seen in sacral giant cell tumors [18]. Nevertheless, giant cell tumors are usually found in patients aged from 20 to 30 years [18]. Multicentric giant cell tumors or multiple bone metastases are also rare [19].

Telangiectatic osteosarcoma fits the description of our patient's sacral tumor. Telangiectatic osteosarcoma is a bone malignancy characterized by multiple, dilated, blood-filled cavities with high-grade sarcomatous cells in the peripheral rim and septations [20]. It usually displays as an osteolytic bone lesion with infiltrative margins on CT images, meanwhile, multiple FFLs with thick peripheral and septal surrounding tissue can be found on MRI [20, 21]. However, patients with telangiectatic osteosarcoma are usually younger than 20 years old [21], and telangiectatic osteosarcoma of the sacrum was extremely rare, with only one case reported in PubMed [22].

In summary, we report an uncommon case of bone metastases with multiple FFLs from gastric cancer, with an atypical appearance of the sacral lesion, which is difficult to be differentiated from a primary sacral malignancy. The point is to know that multiple bone lesions with multiple FFLs can be a sign of bone metastases; meanwhile, the FFLs in bone metastases indicate that breast, gastric, and lung cancers are the most likely primary tumors. In addition, it is important for radiologists to pay attention to the age of the patient and the site of the lesion in order to make a reasonable and accurate diagnosis.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

Informed consent Informed consent was obtained from all individual participants included in the study.

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