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Case report of a rare giant bone island in a vertebral body combined with hemangioma

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Abstract

This case report describes a rare giant bone island combined with hemangioma diagnosed in a patient with osteolytic vertebral metastases. The bone island's greatest diameter was 3.15 cm, and bone islands of this size are rare in the literature. This article aims to provide clinicians with information about the diagnosis and relevant literature of bone islands.

Keywords

Bone island, hemangioma, case report, spine, osteoblastic metastases, radiographic images

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Introduction

Bone islands constitute misplaced normal cortical bone in cancellous bone, and have an incidence rate of one in 1 million.[1] Bone islands occur most often in the femur and tibia and have a general diameter of 0.2 to 2.0 cm, but the spine is rarely involved. Onitsuka reported that only 3 of 209 bone islands (1.4%) occurred in the thoracic and lumbar vertebrae.[2]

On X-ray, bone islands appear as a round or oval hardening shadow in the medullary cavity without periosteal reaction. On computed tomography (CT), bone islands usually appear in the center of the vertebral body, and present with clear boundaries and high density shadows and with a general size of approximately 1 to 2 cm.

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In this report, we describe a rare case of a giant bone island combined with hemangioma in the vertebral body. The purpose was to provide clinicians with information to differentiate between bone islands and osteoblastic metastases.

Case report

The patient was a 63-year-old woman with primary lung cancer. She had been experiencing continuous back pain and limited spinal movement for several days, especially at night. Physical examination showed a specific site of pain and pain on percussion, but no symptoms in either lower extremity. The pain could not be relieved with non-steroidal anti-inflammatory drugs. She underwent CT, magnetic resonance imaging (MRI), and positron emission-computed tomography (PET)-CT. The results showed osteolytic destruction and high F-18 fluorodeoxyglucose (FDG) uptake in several vertebrae; thus, the patient was diagnosed as having multiple vertebral metastases. Unexpectedly, CT also showed high-density shadows in the 7th thoracic vertebra (T7, Figure 1). MRI showed both high- and low signal intensity in T1-weighted/T2-weighted (T1W1\T2W1) images (Figure 2). PET-CT showed fence-like trabecular bone, irregularly dense shadows, and low FDG uptake reduction or defects (Figure 3). The area subsequently

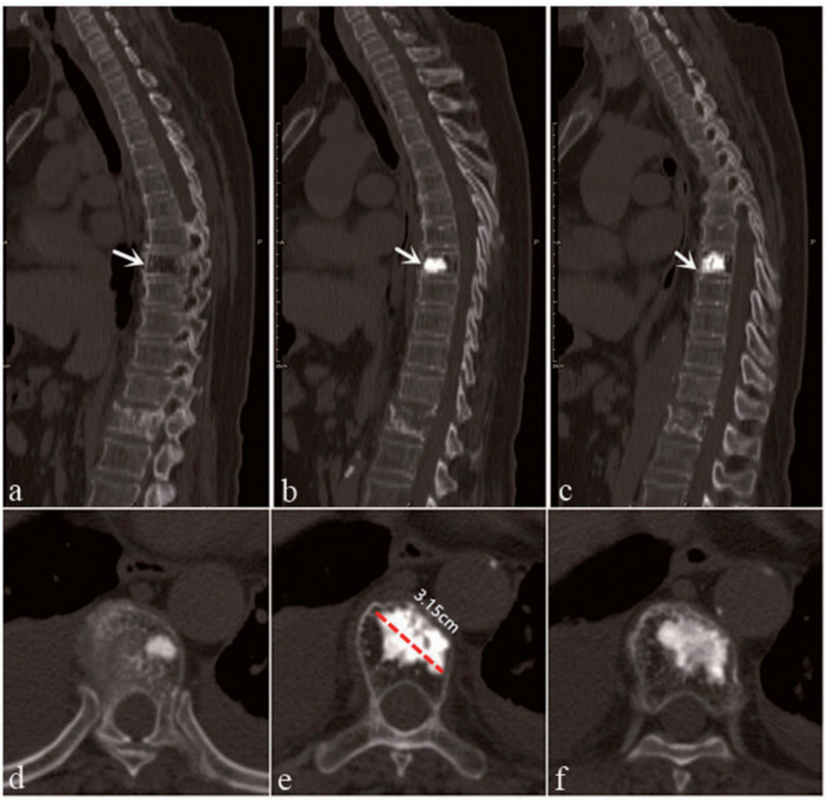

Figure 1. Computed tomography (CT) images. a–e: Low-density shadows combined with high- and uniform-density shadows on sagittal and cross sectional views, which differs from bone metastases, which show bone formation within soft tissue density; e: maximum diameter: 3.15 m.

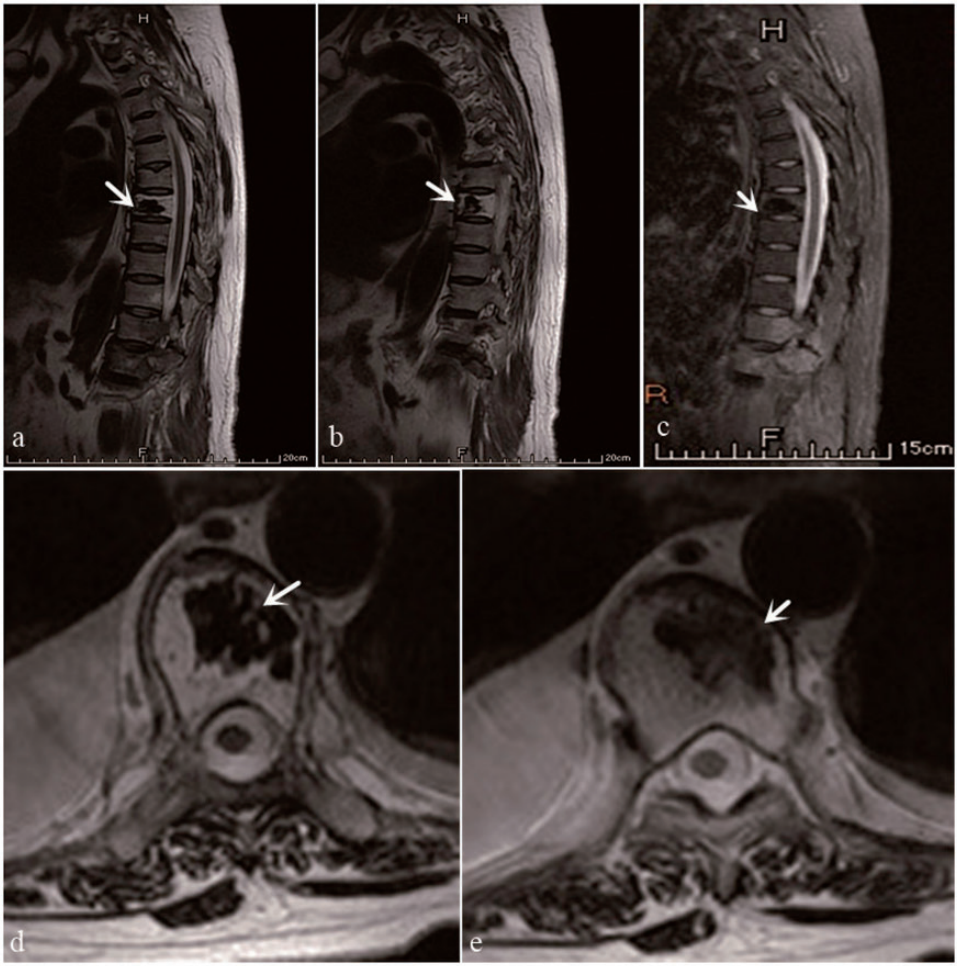

Figure 2. Magnetic resonance images (MRI). a, b, d and e: High signal intensity (hemangioma) combined with low signal intensity (bone island) in T1-weighted/T2-weighted (T1WI\T2WI) images; c: Most of T7 shows a low signal intensity (bone island) in this T2WI fat saturation image, whereas bone metastases show a high signal.

confirmed by histopathology to be a bone island had a maximum diameter of 3.15 cm. To further identify the abnormal presentation in T7, we performed a biopsy while also performing percutaneous vertebroplasty (PVP) for the vertebral metastases (Figure 4a–c). Postoperative pathological analysis identified lamellar bone and abnormal hyperplasia, which is the typical characteristic of a bone island (Figure 4d).

Discussion

In 1979, Smith[3] reported a bone island, described as a giant bone island in the left supra-acetabular area with a diameter of 4 cm. In 1989, Gold et al.[4] described a bone island in the tibia with a diameter of 5 cm. The diameter of the bone island we described was 3.15 cm, and the island appeared as a high-density shadow with

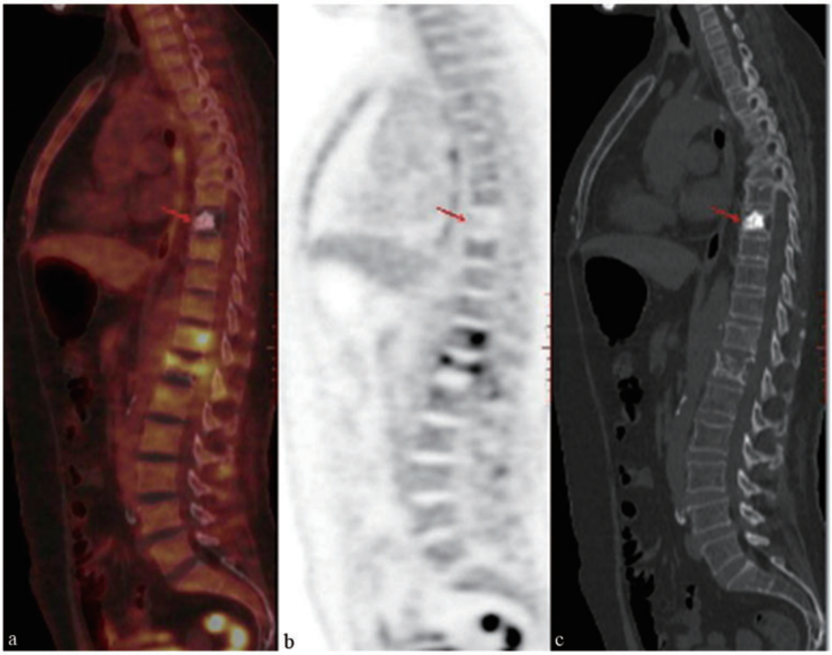

Figure 3. Positron emission tomography-computed tomography (PET-CT) images. a–c: Fence-like trabecular bone, irregular density shadows, and reduced F-18 fluorodeoxyglucose (FDG) uptake or defects (bone island), in contrast to bone metastases, which present with high FDG uptake.

clear boundaries and without spinal fusion. The island was located in the center of the vertebral body on radiographic images (Figure 1–3). These findings distinguish the benign features of a giant bone island.

Bone islands are a benign osteoma and are usually found incidentally because they are asymptomatic. The giant bone island in our patient was found during examination of lumbar vertebral metastasis. Nevertheless, if the bone island is large enough, it may cause continuous pain. Sun et al.[1] reported a giant bone island in the ilium measuring $6.5 \times 7.5\,\text{cm}^2$ that induced continuous dull pain at night. When the giant bone island was excised, the patient's pain was completely relieved. However, how bone islands induce pain requires further study.

One of most important differential diagnoses for bone island is osteogenic metastases. Osteogenic metastases originate as bone-like tissues but proliferate abnormally with incomplete calcium deposition. The presentation is similar to a bone island; however, osteogenic metastases present with a lower density owing to the lower amount of normal cortical bone. The low-density "cancer nest" is a unique characteristic of osteoblastic metastases on multi-slice spiral CT,[5] which is a key differentiating factor from bone island. In addition, bone metastases histologically manifest as bone formation in tumor tissue (soft tissue density on CT), with average, maximum, and minimum CT values lower than those of bone islands.[6]

PET-CT, widely used in tumor diagnosis, plays an important role in identifying bone islands and high-density osteogenic spinal metastases. The metabolism of bone islands is equivalent to that of the

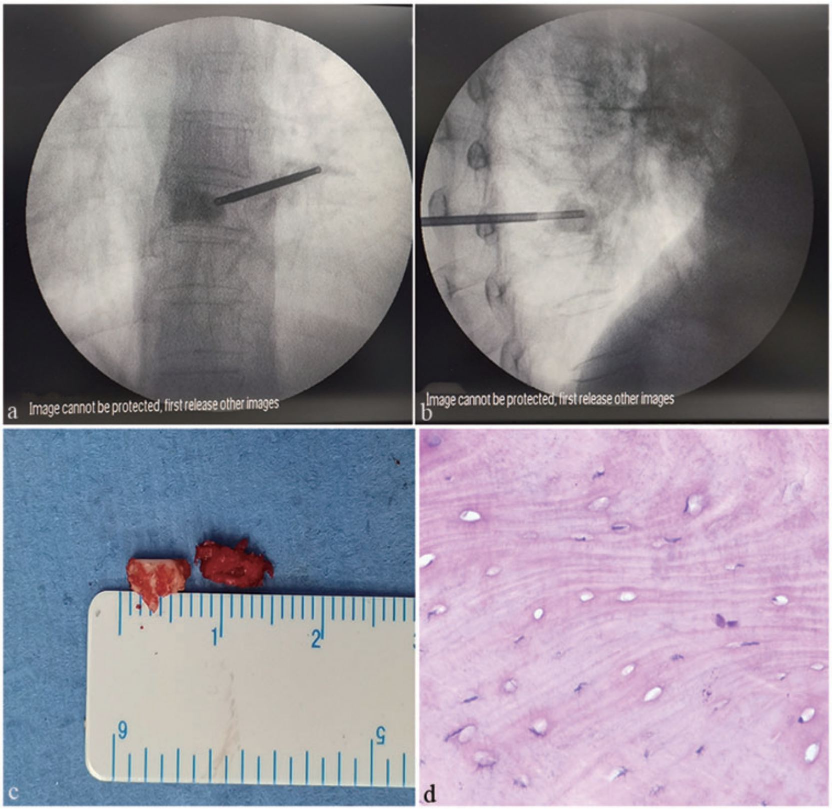

Figure 4. Pathological findings. a, b: Biopsy of T7; c: Two pieces of bone tissue measuring approximately 0.6 cm in size; d: Postoperative pathology showing the typical characteristic of a bone island: abnormally hyperplastic lamellar bone.

surrounding cancellous bone. Bone islands exhibit low FDG uptake on PET-CT, while osteogenic spinal metastases may exhibit high FDG uptake owing to the high metabolism of tumor cells. However, Ran et al.[7] reported a large bone island with high FDG uptake in the left tibia, with a maximum standardized uptake value (SUVmax) of 3.8. The authors concluded that high FDG uptake in bone islands may be caused by hyperproliferative fibroblasts and increased blood flow. In our patient, the bone island showed low FDG uptake, which may have been caused by the minimal bone composition in the vertebral hemangioma, consistent with a previous study.[8]

To achieve an exact diagnosis, bone biopsy is essential to exclude potential malignancy. In the present case, histopathology diagnosed the bone island.

Conclusion

This report described a rare giant bone island combined with hemangioma diagnosed in a patient with osteolytic metastatic vertebral metastases. This case report may provide clinicians with information about the diagnosis and relevant literature of bone island.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethics statement

Ethical approval for this investigation was obtained from the Research Ethics Committee of the First People's Hospital of Longquanyi District (approval No. AK-KY202012). Written consent was obtained from the patient's caregiver.

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