

# Cavernous Hemangioma of the Ilium Mimicking Aggressive Malignant Bone Tumor with Increased Activity on <sup>18</sup>F-FDG PET/CT

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Osseous hemangioma is a benign vascular tumor, and it usually occurs in the vertebrae and the skull. However, hemangiomas of flat bones are rare, and there are very few reports that describe the radiologic findings of osseous hemangioma of the ilium. We report a unique case of large cavernous hemangioma mimicking a chondrogenic malignant bone tumor originated from the ilium in a 22-year-old female. The mass showed stippled calcifications, heterogeneous enhancement with thick septa and enhanced soft tissue components on CT and MR, and also this mass demonstrated heterogeneous 2-fluoro [fluorine-18]-2-deoxy-D-glucose (<sup>18</sup>F-FDG) uptake on <sup>18</sup>F-FDG PET/CT.

**Index terms:** Ilium; Bone tumor; Hemangioma; CT; MR; PET; CT

## INTRODUCTION

Osseous hemangioma is a benign vascular tumor and most frequently involves the bones of the axial skeleton (1, 2). Osseous hemangiomas in the peripheral, tubular and flat bones are uncommon, and very few reports have described the radiologic findings of the osseous hemangioma of the ilium (1-3). Osseous hemangiomas may have a wide variety of radiologic appearances, and the lesions occurred in the tubular or flat bones can be demonstrated as aggressive tumors showing cortical destruction with extraosseous mass formation and osteolytic lesions mimicking a malignant bone tumor (1-6). On 2-fluoro [fluorine-18]-2-deoxy-D-

glucose (<sup>18</sup>F-FDG) positron emission tomography (PET), hemangioma has been classified as a metabolically stable tumor with low <sup>18</sup>F-FDG uptake, but a recently reported case of hemangioma of the tibia showed high <sup>18</sup>F-FDG uptake on PET/CT (7).

We present a case of a cavernous hemangioma of the ilium mimicking an aggressive malignant bone tumor, which demonstrated a large osteolytic lesion with cortical destruction, extraosseous soft tissue mass formation, and irregular calcifications on CT and MR. In addition, we describe the <sup>18</sup>F-FDG PET/CT findings of the osseous hemangioma of the ilium, which showed heterogeneous <sup>18</sup>F-FDG uptake.

## CASE REPORT

A 22-year-old female presented with a gradually increasing pelvic mass with tenderness for 1 month. There was no evidence of previous trauma or fever, and also there was no history of other medical or surgical disease and no evidence of pregnancy. On the physical examination, there was a palpable mass with tenderness in her left pelvic area. A plain radiograph of the pelvis showed a huge, geographic

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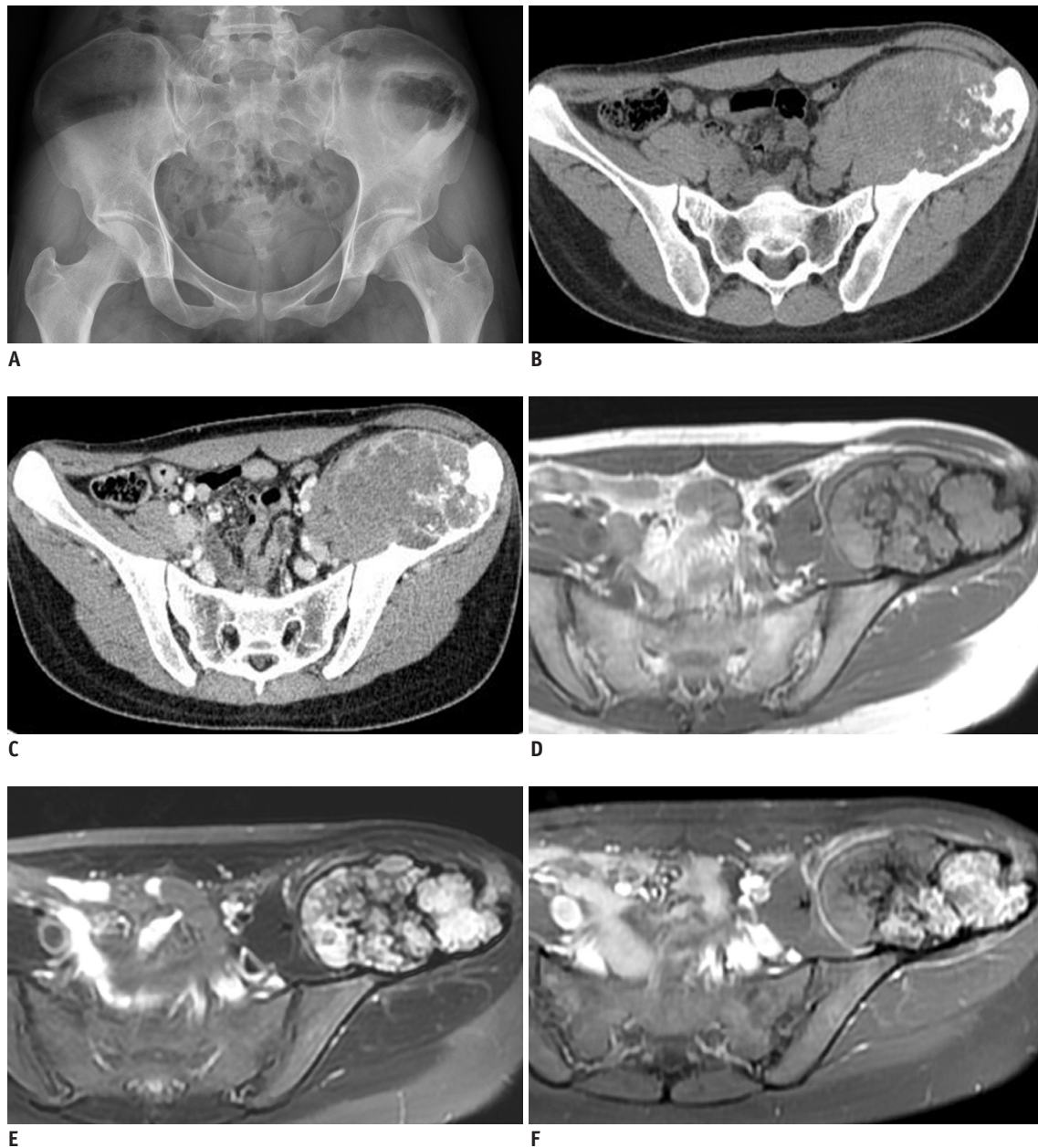
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Cavernous Hemangioma of Ilium Mimicking Aggressive Bone Tumor on FDG PET

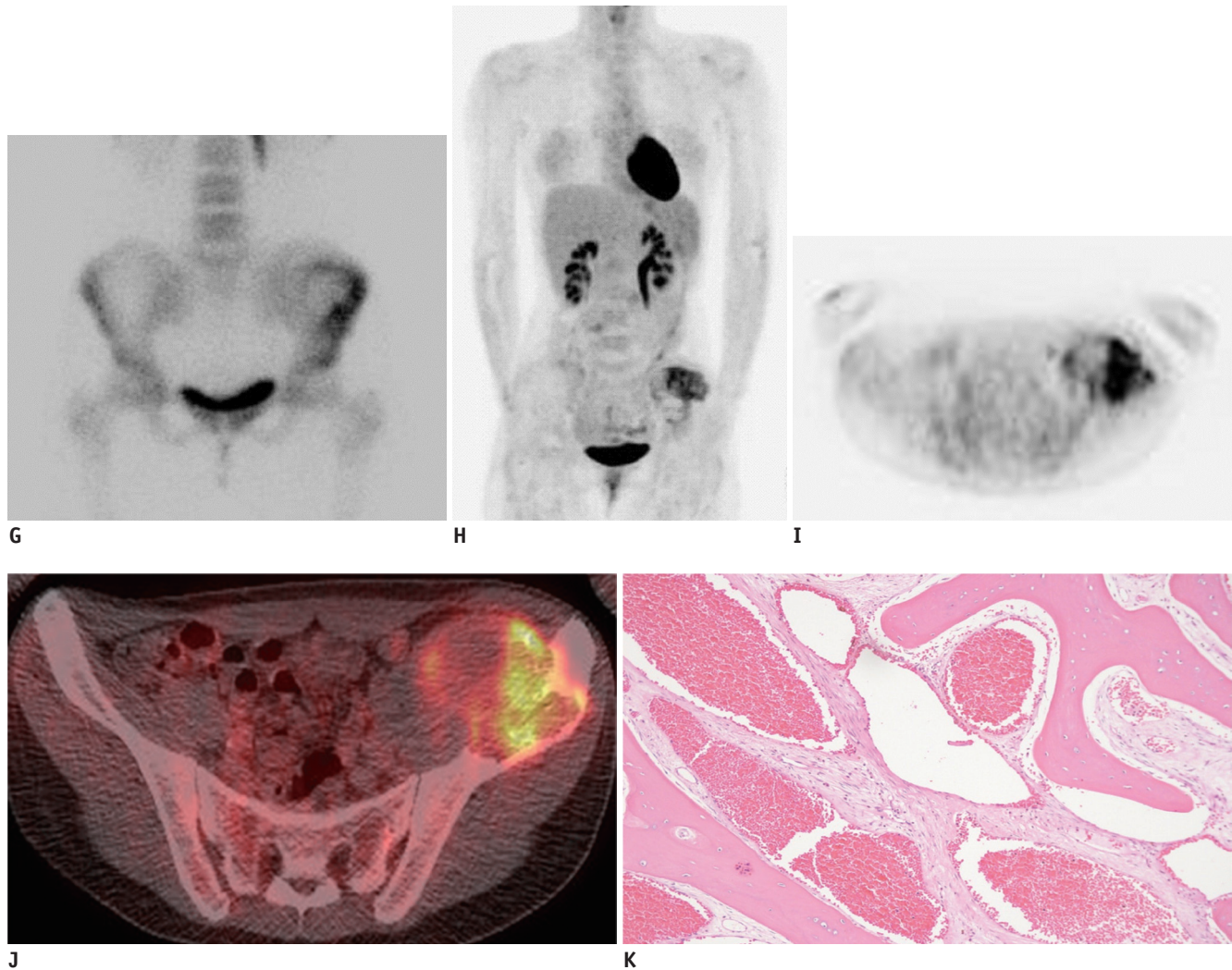
osteolytic lesion with a thick and irregular sclerotic rim in the left ilium (Fig. 1A). CT showed an ill marginated, 13 x 10 cm, osteolytic lesion of the left ilium with extraosseous soft tissue mass formation and intralesional stippled calcifications. On the contrast-enhanced CT, the mass showed irregular, rim enhancement and focal enhanced soft

tissue components (Fig. 1B, C). On MRI, the lesion had a heterogeneous intermediate signal on the T1-weighted images, a heterogeneous high signal on fat suppressed T2-weighted images, and irregular low signal septa on T1 and T2-weighted images. After enhancement with gadolinium, the mass was demonstrated as a heterogeneously enhanced



**Fig. 1. Cavernous hemangioma of ileum in 22-year-old female mimicking aggressive malignant bone tumor with increased activity on <sup>18</sup>F-FDG PET/CT.**

**A.** Plain radiograph of pelvis shows huge, geographic osteolytic lesion with thick, irregular sclerotic rim in left ilium. **B, C.** On precontrast CT image (**B**), there is irregular, large osteolytic lesion in left ilium with exophytic soft tissue mass and stippled calcifications. On contrast-enhanced CT (**C**), mass shows heterogeneous enhancement with irregular, rim and septal enhancement. **D-F.** On MRI, mass has heterogeneous intermediate signal on T1-weighted image (**D**), heterogeneous high signal on fat suppressed T2-weighted image (**E**), and thick irregular low signal septa on T1 and T2-weighted images. After enhancement with gadolinium, mass shows heterogeneous enhancement and non-enhanced septa (**F**). <sup>18</sup>F-FDG = 2-fluoro [fluoroine-18]-2-deoxy-D-glucose



**Fig. 1. Cavernous hemangioma of ileum in 22-year-old female mimicking aggressive malignant bone tumor with increased activity on  $^{18}\text{F}$ -FDG PET/CT.**

**G.**  $^{99\text{m}}\text{Tc}$  hydroxymethane diphosphonate bone scan. Anterior view shows increased peripheral isotope uptake of left iliac bone lesion. **H-J.**  $^{18}\text{F}$ -FDG PET MIP (**H**), PET axial (**I**), and PET/CT fusion axial (**J**) images show heterogeneously increased FDG uptake of left iliac bone lesion (maximum SUV = 5.1). **K.** Photomicrograph shows dilated blood-filled vessels lined by flattened endothelial cells (H/E, x 200).

mass, as well as non-enhanced soft tissue components and septal structures (Fig. 1D-F). On  $^{99\text{m}}\text{Tc}$  hydroxymethane diphosphonate (HDP) bone scintigraphy, increased isotope uptake was depicted in the left iliac bone (Fig. 1G). After fasting for 6 hours, intravenous injection of 375 MBq of  $^{18}\text{F}$ -FDG was performed (at that time, her blood glucose level was 92 mg/dL). The PET scan (3 min/bed, 6-8 beds) was obtained from 60 minutes post  $^{18}\text{F}$ -FDG injection.  $^{18}\text{F}$ -FDG PET/CT scanning was done by Discovery PET/CT scanner (GE Healthcare, Milwaukee, WI, USA), and scans were acquired in 3D mode. Reconstruction was done by ordered subset expectation maximization (OSEM) (128 x 128 matrix, 3.27 mm slice thickness, subsets: 21, iteration: 2, post-Filter: 5.14 FWHM (mm), loop-Filter: 4.69 FWHM, diameter: 60 cm)

after attenuation correction.  $^{18}\text{F}$ -FDG PET/CT demonstrated heterogeneous  $^{18}\text{F}$ -FDG uptakes, which had low and high  $^{18}\text{F}$ -FDG uptake (maximum standard uptake value [ $\text{SUV}_{\text{max}}$ ] = 5.1) within the mass (Fig. 1H-J). Based on the imaging features, chondrogenic malignant bone tumor in the ilium was considered even though, when taking her age into account, it was unusual. Complete removal of the tumor was performed. On gross examination, the mass appeared as a vascular mass containing central necrosis, irregular bony septa, and fibrous tissues. Histopathologic examination of the excised mass showed bony trabeculae with dilated vascular spaces filled with blood and endothelial lining (Fig. 1K), and it was confirmed as cavernous hemangioma of the ilium by the experienced musculoskeletal pathologists.

## DISCUSSION

Intraosseous hemangiomas are benign hamartomatous vascular tumors of the bones, and they frequently involve the skull and vertebral column (1-3). The peripheral, long, tubular and flat bones are the least affected sites, and among them, the tibia and femur are the most common sites (1). Very few cases of hemangioma affecting the pelvic bones have been reported (1-3). Hemangiomas involving the vertebrae or skull are usually asymptomatic and they are often found incidentally on routine imaging studies. However, hemangiomas in the peripheral and pelvic bones are usually symptomatically associated with pain, soft tissue swelling and pathologic fracture (1, 2, 5). Our patient complained of tenderness with a palpable mass in her pelvis for a duration of approximately one-month.

Intraosseous hemangiomas may have various radiologic findings. Due to the diversity of radiologic features of this disease, a correct preoperative diagnosis is rarely made. On plain radiographs, osseous hemangiomas may show a loculated, sunburst, moth-eaten, and soap bubble appearance due to engorged vessels and thickened trabeculae, and they rarely manifest expansile osteolytic changes (1-8). On CT, hemangiomas in the skull and spine can present a typical polka-dot or honeycomb appearance; however CT findings of the hemangiomas in long and flat bones are non-specific and it is difficult to take into consideration osseous hemangiomas in a differential diagnosis. They can be demonstrated as expanded lytic areas surrounded by coarse trabecular bone with or without a soft tissue component, a lesion with a "polka-dot" pattern, an osteoplastic lesion with a sun-burst-like pattern, honeycombed appearance with cortical defects, an irregular destructive lesion with scalloping on the inner surface of the sclerotic cortical bone, and occasionally calcifications have been reported (1-4, 6, 8, 9). In our case, the mass was depicted as an osteolytic lesion with cortical destruction and extraosseous soft tissue mass formation, stippled calcifications on CT, and enhanced, exophytic soft tissue components on contrast-enhanced CT mimicking chondrogenic malignant bone tumors.

On MR, osseous hemangiomas usually show a variety of signals from low to very high signal intensities on T1- and T2-weighted images within the lesion according to the internal components such as dilated vessels, hemorrhage, blood clots, fibrosis, bony trabeculations, fat, and smooth muscles (1-7). Osseous hemangiomas can show variable

degrees of enhancement related to internal contents, as we mentioned above, after contrast administration. Low signal septal structures within the lesion on T1 and T2-weighted images, and the appearance of a bunch of grapes on T2-weighted and Gd-enhanced images were also reported (3, 6-8). In our case, non-enhanced, thick, irregular, septa like structures of low signal intensity within the mass were demonstrated on MR, and we retrospectively thought that this finding could be perceived as a soap-bubble appearance lesion that can be shown in the usual osseous hemangiomas.

$^{18}\text{F}$ -FDG PET/CT is increasingly being used to obtain more information for differentiating benign and malignant tumors. Even though, there are some false positive and false negative  $^{18}\text{F}$ -FDG uptakes in benign and malignant bone tumors, respectively, PET is generally used in the diagnostic work-up of various musculoskeletal tumors. Hemangioma has been known to be a metabolically stable benign lesion on PET. In vertebral hemangioma, it has been reported to demonstrate from ametabolic, cold vertebra to low  $^{18}\text{F}$ -FDG uptake on PET/CT images. This finding can help to differentiate a hemangioma from a metastatic lesion in the case of a vertebral hemangioma depicted as a photopenic defect at the  $^{99\text{m}}\text{Tc}$  MDP bone scan, which need to be differentiated from osteolytic metastatic lesions in patients with a malignant disease. (10, 11). Hatayama et al. (12) have described the SUVs for  $^{18}\text{F}$ -FDG in 16 hemangiomas as ranging from 0.73 to 1.67, and Tian et al. (13) have described the SUVs for  $^{18}\text{F}$ -FDG in 4 hemangiomas as ranging from 1.6 to 2.3. However, Cha et al. (7) recently reported the case of an intra-osseous hemangioma of the tibia which showed increased  $^{18}\text{F}$ -FDG uptake (SUV = 3.9) on PET/CT. Similar to the case reported from Cha et al. (7), our case of the osseous hemangioma of the ilium showed heterogeneously increased  $^{18}\text{F}$ -FDG uptake (SUV<sub>max</sub> = 5.1) on PET/CT. Sakurai et al. (14), and Higashiyama et al. (15) had described that  $^{18}\text{F}$ -FDG PET may provide useful information to differentiate hemangioma from malignant soft tissue tumors owing to the tendency of low  $^{18}\text{F}$ -FDG uptake in soft tissue hemangiomas. However, contrary to soft tissue and vertebral hemangiomas, osseous hemangiomas in the peripheral and flat bones can show high  $^{18}\text{F}$ -FDG uptake on PET/CT as in our case and the case from Cha et al. (7). To our knowledge, there was no radiologic report describing osseous hemangiomas of the ilium which demonstrated an increased  $^{18}\text{F}$ -FDG uptake on PET/CT.

Intraosseous hemangiomas of the long bones and ribs

can be rarely demonstrated as an aggressive lesion showing extraosseous growth with the disrupted bony cortex (1-4, 8). The differential diagnosis for a hemangioma of long bones included a giant cell tumor, aneurysmal bone cyst, fibrous dysplasia, and plasmacytoma (1, 5). In the case of hemangiomas of the flat bones which show aggressive growth and osteolytic lesions, malignant bone tumors can be considered as a primary differential diagnosis (1, 3-5). In our case, chondrosarcoma was the primary differential diagnosis although the incidence of the chondrosarcoma is rare for her age.

In conclusion, cavernous hemangioma of the ilium can show cortical destruction, stippled calcifications, and an extraosseous soft tissue mass formation, which can give the false impression of a chondrosarcoma. Furthermore, osseous hemangiomas in the peripheral and flat bones can demonstrate high <sup>18</sup>F-FDG uptake on PET/CT. Although osseous hemangioma of the ilium is rare, when a young patient presented with an osteolytic lesion with an extraosseous soft tissue mass, stippled calcifications and thick septa or coarse trabeculations, osseous hemangioma should be taken into consideration in the differential diagnosis. However, the final diagnosis can only be confirmed by histopathologic examination.

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