CASE REPORT

PET/CT and MRI of intra-osseous haemangioma of the tibia

¹J G CHA, MD, ²J H YOO, MD, ³H K KIM, MD, ⁴J M PARK, MD, ¹S H PAIK, MD and ⁵S J PARK, MD¹Department of Radiology, Soonchunhyang University Bucheon Hospital, Bucheonsi, Gyeonggido, Republic of Korea,²Department of Orthopedics, Seoul SKY Hospital, Seoul, Republic of Korea, ³Department of Pathology, Soonchunhyang University Bucheon Hospital, Bucheonsi, Gyunggi-do, Republic of Korea, ⁴Department of Nuclear Medicine, Soonchunhyang University Bucheon Hospital, Bucheonsi, Gyunggi-do, Republic of Korea, and ⁵Department of Radiology, Kyung-hee University Hospital, Dongdaemun-gu, Seoul, Republic of Korea

ABSTRACT. Intra-osseous haemangioma is a rare, benign neoplasm that usually involves the vertebrae and craniofacial bones. Furthermore, its occurrence in the long bones is extremely rare. We report the findings of fluorine-18-fludeoxyglucose (¹⁸F-FDG) positron emission tomography (PET)/CT and MRI in a patient with intra-osseous haemangioma in the proximal tibia, who was initially misdiagnosed as having a malignancy based on ¹⁸F-FDG PET/CT. ¹⁸F-FDG PET/CT showed a well-marginated osteolytic lesion with abnormal FDG uptake. The mass demonstrated low signal intensity on T_1 weighted MRI. On T_2 weighted images, the lesion appeared as a cluster of high signal intensity lobules and showed strong enhancement on contrast-enhanced T_1 weighted images. Surgical curettage was performed and histopathological examination of the excised tissue confirmed a cavernous haemangioma.

Received 9 March 2011

Revised 13 June 2011

Accepted 20 July 2011

DOI: 10.1259/bjr/35251836

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Intra-osseous haemangioma is a rare benign neoplasm that usually involves the vertebrae and craniofacial bones [1, 2]. Radiological findings of this tumour in the skull and spine are so typically characteristic that radiologists can differentiate it from other bone tumours [1]. In contrast to reported cases of intra-osseous haemangioma with typical clinical presentations, the correct diagnosis of this tumour when it occurs in the long bones or shows atypical radiological findings may be more challenging for musculoskeletal radiologists and orthopaedic surgeons, leading to unnecessary clinical and radiological studies.

Previous studies have described positron emission tomography (PET)/CT imaging as an accurate method for pre-operative staging of bone and soft-tissue sarcoma [3–5]. However, the use of PET imaging in the differential diagnosis between benign and malignant bone tumours has been debated because high accumulation of fludeoxyglucose (FDG) can be observed in some benign bone lesions, especially histiocytic or giant cell containing lesions [6]. Previous studies have reported that the maximal standardised uptake value (SUV_{max}) of haemangioma was less than 2.5 on fluorine-18-FDG (¹⁸F-FDG) PET/CT [6–9]. To our knowledge, these atypical ¹⁸F-FDG PET/CT findings of intra-osseous haemangioma have not been previously reported. We report a case of intra-osseous haemangioma occurring in the tibia that mimicked a malignancy and describe CT, MRI and ¹⁸F-FDG PET/CT findings.

Case report

A 73-year-old male was admitted to our hospital for a complete medical evaluation of a lesion with abnormal FDG uptake in the right proximal tibia that was found on an ¹⁸F-FDG PET/CT scan during his annual check-up. The ¹⁸F-FDG PET/CT (Figure 1a–c) showed abnormal uptake [maximum standardised uptake value (SUV_{max})=3.9] in the osteolytic lesion of the right proximal tibia. A physical examination revealed no evidence of tenderness or a palpable mass around either knee joint. The range of motion of both knee joints was within normal limits. The radiographs of the right knee joint demonstrated no bony abnormality or joint space narrowing (Figure 2a,b).

An MRI exam was performed using a 3.0 T scanner. T_1 weighted images (Figure 3a) showed a low signal intensity mass lesion located in the epimetaphysis of the medial condyle of the right tibia. On T_2 weighted images (Figure 3b), the mass lesion appeared as a cluster of multiple high signal intensity lobules with multiple septations containing several round areas of low signal intensity. Contrast-enhanced T_1 weighted images (Figure 3c,d) showed strong enhancement of the mass, but no enhancement was observed in the areas that demonstrated low signal intensity on T_2 weighted images. The initial radiological diagnosis included metastasis, plasmacytoma, giant cell tumour and clear cell chondrosarcoma. The patient underwent curettage followed by cement insertion. Microscopic analysis revealed large, blood-filled vascular channels that were lined with thin, flattened epithelium (Figure 4a). No evidence of malignancy was found. Immunohistochemistry for CD34 expression revealed a positive result in the lining endothelial cells (Figure 4b). The final diagnosis was a cavernous haemangioma of the proximal tibia.

Address correspondence to: Dr Jang-Gyu Cha, Department of Radiology, Soonchunhyang University Bucheon Hospital, 1174 Jung-Dong, Wonmi-Gu, Bucheon, Gyeonggi 420-853, Republic of Korea. E-mail: mj4907@schmc.ac.kr

Figure 1. Fluorine-18-fludeoxyglucose (^{18}F -FDG) positron emission tomography (PET)/CT image. (a) CT image shows a geographic osteolytic lesion in the right proximal tibia (arrow). (b) On an integrated ^{18}F -FDG PET/CT image, the lesion shows abnormal FDG uptake (maximal standardised uptake value=3.9). (c) Coronal PET maximum intensity projection shows a lesion with high uptake (arrow) on the medial side of the right proximal tibia.

Discussion

Intra-osseous haemangioma is a rare bone tumour, accounting for <1% of all bone neoplasms [10]. Additionally, its occurrence in the long bones is extremely rare and it can develop in patients of any age group, but is most commonly found in 30- to 40-year-old patients [11]. Women are affected twice as often as men. These tumours typically present as a solitary mass, although diffuse bone involvement has been documented [12].

Unlike haemangioma occurring in the skull and spine, most haemangioma involving the extra-axial

skeleton cause clinical symptoms, such as local pain and swelling around the lesions [13]. However, in our case the tumour was found incidentally during a complete medical check-up, which can be explained by the fact that, in many medical institutions in South Korea, PET CT imaging is an option in comprehensive medical check-up programmes for the evaluation of hidden malignancy.

Including our case, we found only five examples in the literature in which the tumour originated from the extra-axial skeleton and initially presented as incidental radiological findings.

Figure 2 (a) Anteroposterior and (b) lateral radiographs of the right knee joint demonstrate no abnormal lesion in the corresponding area of the right tibia.

Figure 3. MR images. (a) The sagittal T_1 weighted image [repetition time/echo time (TR/TE), 467/10] shows a low signal intensity mass lesion involving the epimetaphysis of the right proximal tibia. (b) On the sagittal T_2 weighted image (TR/TE, 4000/73), the mass lesion appears as a cluster of high signal intensity lobules, resembling a bunch of grapes or a honeycomb. The thrombus is observed (arrow) as areas of low signal intensity within the mass. (c) The sagittal contrast-enhanced T_1 weighted image (TR/TE, 800/12) reveals homogeneous enhancement in the mass, but the thrombus (arrow) in the mass showed no enhancement. (d) The coronal contrast-enhanced T_1 weighted image (TR/TE, 533/12) shows a strong enhancing mass lesion centred around the tibial tuberosity with multiple thrombi (arrow).

Histologically, haemangioma can be classified as cavernous, capillary, venous or mixed, depending on the type of vascular involvement [13]. Cavernous haemangioma is the most common type of intra-osseous haemangioma arising from extremity bone and constitutes 50% of all reported cases. They are typically located at the medullary and intracortical portions of the bone [13]. Pure capillary haemangioma constitutes 10% of all the types reported in the literature [14]. Venous haemangioma has been rarely reported in extremity bone [13]. Haemangioma may be observed as well-defined, primarily osteolytic, variably expansile lesions with a radiating lattice-like or web-like coarse trabecular pattern similar to those of haemangioma in the skull or spine [11]. The MRI findings of intra-osseous haemangioma may vary depending on the proportion of vascular and lipomatous soft-tissue elements [15]. When the tumour is composed primarily of vascular structures, it shows low signal intensity on T_1 weighted images and high signal intensity on short tau inversion recovery and T_2 weighted images, probably owing to slow blood flow [2]. However, as the amount of the fat component in the tumour increases, the signal intensity on T_1 weighted and T_2 weighted images tends to increase [1]. Contrast-enhanced MRI shows variable degrees of enhancement [1, 13]. Vilanova et al [2] described that on T_2 weighted images, soft-tissue haemangioma may present as a cluster of multiple high signal intensity lobules that resemble a "bunch of grapes", resulting from cavernous or cystic vascular spaces

containing stagnant blood. Additionally, areas of thrombosis in haemangioma are seen as low signal intensity areas surrounded by a high signal rim on T_2 weighted images [2]. The authors suggested that these MRI features may be diagnostic, particularly when the mass is deep seated. In our case, the tumour also demonstrated a bunch of grapes appearance as well as areas of thrombosis on T_2 weighted and contrast-enhanced T_1 weighted images. To our knowledge, these MRI findings in intra-osseous haemangioma have not been documented in the English-language medical literature.

Although ^{18}F -FDG PET/CT is increasingly being used in the diagnostic work-up of various tumours that are suspected of being malignant, the clinical use of ^{18}F -FDG PET to differentiate between malignant and benign musculoskeletal tumours has been controversial because high ^{18}F -FDG uptake has been detected in some benign tumours and low uptake has been detected in some malignant tumours [5, 9]. Benign lesions that are recognised as showing false-positive FDG uptake ($\text{SUV}_{\text{max}}=2.0$) include pigmented villonodular synovitis, sarcoidosis, hibernoma, neurofibroma, schwannoma, desmoid fibromatosis, giant cell tumour, osteoid osteoma, histiocytosis-X, chondroblastoma, enchondroma and non-ossifying fibroma [5–7, 16]. However, haemangioma has been classified as a metabolically stable benign tumour on PET imaging [6–8]. Hatayama et al [8] have described the SUVs for FDG in 16 haemangioma as ranging from 0.73 to 1.67, including soft-tissue and osseous ones. They suggested that

Figure 4. Photographs of pathological specimens. (a) Histology of the curettage specimen reveals large, blood-filled vascular channels (haematoxylin and eosin staining; original magnification, $\times 20$). (b) Immunohistochemistry for cluster of differentiation 34 expression shows positivity in the lining endothelial cells (haematoxylin and eosin staining; original magnification, $\times 40$).

FDG PET may provide a useful indicator for differentiation of haemangioma from malignant soft-tissue sarcoma. In our case, the SUV_{max} was 3.9 on an 18F-FDG PET scan, which is about two times higher than the previously suggested cut-off point ($SUV_{max}=2.0$) [17], suggesting that the tumour was malignant in nature. To our knowledge, such a high value has not been previously reported.

Although pre-operative biopsy provides valuable information for optimal surgical planning [18, 19], in our case, pre-operative biopsy was not performed because the patient refused.

The thin-walled blood vessels of haemangioma may be disrupted in the process of curettage or needle biopsy, causing histological sections to show non-diagnostic empty spaces with scattered bone trabeculae. This is the reason why a diagnosis based on biopsy specimens can be challenging to the pathologist [13].

Although special treatment is not required for asymptomatic and small tumours, surgical treatment such as curettage or complete surgical resection and bone grafting is indicated for symptomatic ones. Radiation therapy to ablate the venous channels forming the lesion is reserved for subtotal resection or for an unresectable lesion in a symptomatic patient [1, 20]. Unlike haemangioma occurring in the skull or spine, which have typical radiological features, the diagnosis of extremity-based haemangioma may be more challenging for musculoskeletal radiologists and orthopaedic surgeons. This is because they are rare and lack characteristic radiological findings which may, as in our case, lead to unnecessary clinical and radiological studies. In our case, the tumour presented with abnormal FDG uptake on PET/CT scans across the epiphysis around the knee joint, which led to confusion with other disease entities, including metastasis, plasmacytoma, clear cell chondrosarcoma and giant cell tumour.

Conclusion

Although rare, radiologists should be aware that intra-osseous haemangioma has the potential to demonstrate

abnormal uptake on FDG PET, which could lead to false-positive interpretation. Characteristic MRI findings such as a cluster of multiple high signal intensity lobules and areas of thrombosis may be helpful in the correct diagnosis of the intra-osseous tumour arising from extremity bone.

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