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Chondromyxoid Fibroma of the Femur Can Not Be Differentiated from Chondrosarcoma by 18F-FDG PET/CT, and Histopathology is Still the Final Diagnostic Tool

Taishi Okada, Hiroyuki Futani*, Ryo Kanto, Shunsuke Kumanishi, Yoshitane Tsukamoto and Shinichi Yoshiya

Department of Orthopaedic Surgery, Hyogo College of Medicine, Nishinomiya, Hyogo, Japan

Abstract

Chondromyxoid fibroma is an extremely rare, benign cartilaginous tumor, which might be misdiagnosed as hondrosarcoma. Recent studies reported that PET/CT could distinguish benign cartilaginous tumors from chondrosarcomas with maximum Standardized Uptake Value (SUVmax) of more than 2.0. In the literature, 4 cases of chondromyxoid fibroma have been reported on PET/CT with high accumulation of 18FFDG. However, no paper has explained the reason for this high accumulation. In this paper, we present a case of femoral chondromyxoid fibroma and discuss the rational reason for high accumulation of 18F-FDG by PET/CT in accordance with histology. Here, a 20-year-old female presented with a lesion located in the medial aspect of the left distal femur. Radiography revealed an eccentric radiolucency in the metaphysis of the left distal femur. CT images clearly demonstrated a cortical destruction of the posterior wall. PET/CT images clearly demonstrated an abnormal 18F-FDG uptake of the distal aspect of the left femur with SUVmax value of 6.6, indicating a chondrosarcoma. In the present case, histology showed a number of multinucleated giant cells at the periphery of the lobules in the tumor, which can explain the high accumulation.

Keywords: Chondromyxoid fibroma; PET/CT; Imaging; Chondrosarcoma; Femur

Introduction

The Chondromyxoid Fibroma (CMF) is an extremely rare, benign cartilaginous tumor of the bones, which constitutes less than 0.5% of all primary bone tumors [1,2]. Although it is a benign condition, this tumor has a potentially aggressive behavior with regional expansion. Therefore, CMF can be difficult to differentiate from chondrosarcomas since these 2 conditions overlap by imaging findings [3,4] and even by histology [1,2,5-7].

Recently, the usefulness of fluorine-18 fluorodeoxyglucose (18F-FDG) Positron Emission Tomography (PET)/Computerized Tomography (CT) has been reported in diagnosing malignant cartilaginous tumors, particularly those with borderline clinical, histological and imaging characteristics [8-11]. Feldman et al. [8] reported that the cutoff value of 2.0 of maximum standardized uptake value (SUVmax) could distinguish benign from malignant cartilaginous tumors. To date, only 4 cases of CMFs have been reported on 18F-FDG PET/CT of chondromyxoid fibromas with detailed information [12-15]. Interestingly, those 4 cases showed a high accumulation of 18F-FDG with PET/CT. However, no paper has explained the reason for this high accumulation of 18F-FDG.

Here, we describe a case with femoral CMF, which could be evaluated by 18F-FDG PET/CT. Furthermore, we discuss the rational reason for a high accumulation of 18F-FDG of CMF in relation to the histology.

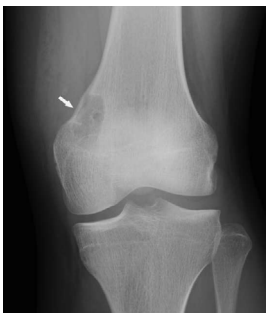

Figure 1: An anterior X-ray shows an eccentric radiolucency in the medial metaphysis of the left distal femur (arrow).

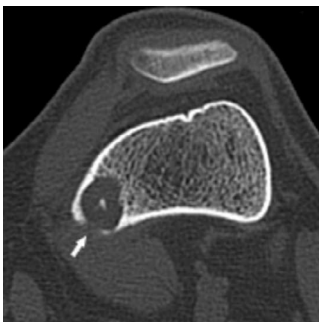

Figure 2: An axial CT image shows an expanding osteolytic lesion with fine calcification. The posterior cortical wall is thin and partly destroyed due to tumor expansion (arrow).

Case Report

A 20-year-old female presented with a lesion located in the medial aspect of the left distal femur. The lesion was unexpectedly found by radiography taken at regional hospital when she noted the left knee pain after stumbling in a stair. Physical examination showed tenderness in the lateral aspect of the metaphysis in the distal femur. Radiography revealed an eccentric radiolucency in the metaphysis of the left distal femur (Figure 1). CT images clearly demonstrated an expanding osteolytic lesion with fine calcification. In addition, a cortical destruction of the posterior wall was also found (Figure 2). Magnetic Resonance Imaging (MRI) revealed a 2.8 × 2.5 × 1.8 cm well-defined mass. An area

***Corresponding author:** Hiroyuki Futani, Department of Orthopaedic Surgery, Hyogo College of Medicine, Nishinomiya, Hyogo, Japan, Tel: + 81 798 45 6452; Fax: + 81 798 45 6453; E-mail: h-futani@hyo-med.ac.jp

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of low signal intensity was found by T1-weighted images (Figure 3). On T2-weighted images, the tumor had a high signal intensity area including a low signal intensity area, which locked like a target (Figure 4). Those findings indicated a cartilaginous tumor. Furthermore, PET/CT images clearly demonstrated an abnormal 18F-FDG uptake of the distal aspect of the left femur with SUVmax value of 6.6, indicating a chondrosarcoma (Figure 5). No evidence of distant metastases was found.

A needle biopsy was performed. The histology of the specimen showed chondromyxoid fibroma or grade II chondrosarcoma. En bloc resection of the tumor was performed with a wide margin followed by internal fixation and polymethylmethacrylate (Surgical Simplex P; Stryker Howmedica Osteonics, Allendale, NJ, USA) filling. The histology of the resected sample showed a lobulated growth pattern. The lobules consisted of spindle cells with a myxoid background (Figure 6). The periphery of the lobules was hypercellular with multinucleated giant cells (Figure 7). Based upon the histological findings, the tumor was diagnosed as a CMF.

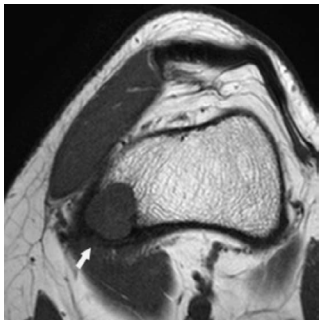

Figure 3: An axial view of a T1-weighted image shows an area of low signal intensity (arrow).

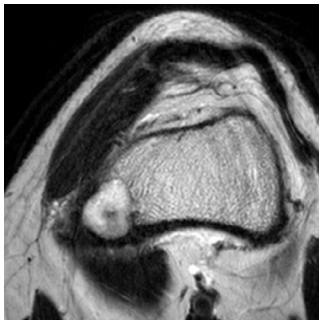

Figure 4: An axial view of a T2-weighted image shows the tumor with a high signal intensity area including a low signal intensity area.

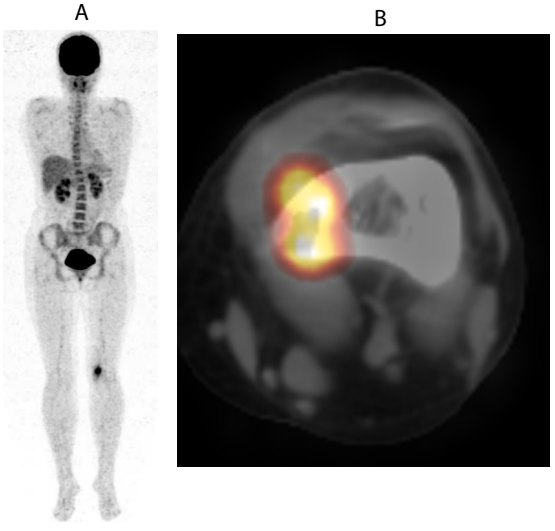

Figure 5: An anterior (A) and an axial (B) view of 18F-FDG PET/CT images reveal an increased uptake of the distal aspect of the left femur (SUVmas 6.6).

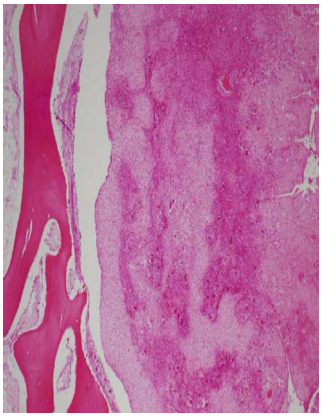

Figure 6: Histology of the tumor consists of lobules with spindle cells and a myxoid background (hematoxylin and eosin staining, original magnification × 20).

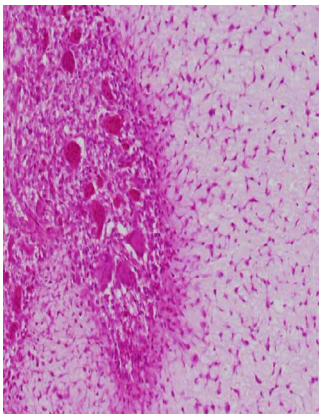

Figure 7: Histology of the tumor consists of multinucleated giant cells in a hypercellular the periphery of the lobules (hematoxylin and eosin staining, original magnification ×100).

The patient started walking on the next day after surgery. Two weeks postoperatively, the knee regained normal range of motion. No recurrence has been found 2 years after surgery (Figure 8).

Discussion

CMF is a benign but locally aggressive tumor occurring primarily in young patients between the second and third decades of life [1,5,16-19]. The most common location is the metaphysis of long tubular bones, especially proximal tibia. Wu et al. [5] reported that among 277 cases with CMF, 130 cases (46.9%) involved the long bones, followed by 84 cases (30.3%) the flat bones, 48 cases (17.3%) the bones of the hands and feet, and 15 cases (5.4%) the skull and facial bones. In their series, 20 cases (7%) were found in the distal femur.

It is important to distinguish CMF from chondrosarcoma because their natural history and prognosis are different. The typical radiological finding of CMF is the presence of a lobular, eccentric, and

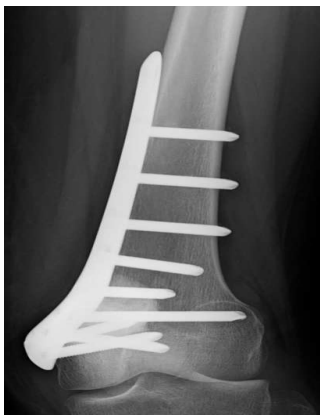

Figure 8: An anterior X-ray taken 2 years after surgery reveals no area of radiolucency around the polymethylmethacrylate.

osteolytic lesion with occasionally expansion of the affected bone [1]. CMF might be aggressive since cortical thinning and expansion are also very common features, and complete cortical destruction may be seen in almost one third of cases [16]. Therefore, the literature emphasized the danger of mistaking this condition for a chondrosarcoma by radiography and/or CT images [3], MRI [4], and sometime even by histology [1,2,5-7].

In distinguishing CMF from chondrosarcoma, the existence of calcification might be useful since calcification is rare in CMFs but common in chondrosarcomas [7]. In the present case, calcification was found in addition to a defect of the cortical bone, suggesting a chondrosarcoma. 18F-FDG PET/CT is a diagnostic imaging technique to detect glucose uptake by cells with high metabolic activity, such as heart, brain, and tumor cells. Recent reports have mentioned that 18F-FDG PET is a useful method to differentiate between benign and malignant cartilaginous tumors [8-10]. The cutoff value of 2.0 of SUVmax could distinguish benign from malignant cartilaginous tumors with overall sensitivity 90.9%, specificity 100%, and accuracy 96.6%, respectively [8]. Lee et al. [9] reported that grade 1 chondrosarcoma is difficult to differentiate from chondroma. However, the cutoff for SUVmax of 2.3 was useful to differentiate grade II and III chondrosarcomas from benign cartilaginous tumor and grade I chondrosarcomas. The positive predictive value was 0.82 (95% confidence interval, 0.48 to 0.97) and the negative predictive value was 0.96 (95% confidence interval, 0.77 to 1.00).

Even though CMF is a benign tumor, the previous reported 4 cases of CMFs showed a high accumulation of 18F-FDG by PET/CT with SUVmax ranging from 3 to 5.9 [12-15]. In the present case, we found the highest accumulation of 18F-FDG with SUVmax of 6.6 in comparison to the previous cases. Aoki et al., (10] reported that a high accumulation of 18F-FDG could be caused by histiocytic or multinucleated giant cells found in lesions such as giant cell tumor, chondroblastoma, Langerhans cell histiocytosis, fibrous dysplasia, and sarcoidosis. Multinucleated giant cells, such as macrophages, play a central role in the host response to injury and infection, and their energy is predominantly supplied by means of intracellular glucose metabolism [17,18]. Schajowicz et al., [2] mentioned that multinucleated giant cells were almost a constant histological finding of CMF in 31 out of 32 cases (97%). In the present case, histology also showed a number of multinucleated giant cells at the periphery of the lobules in the tumor, which can be the reason for high accumulation of FDG with PET.

In the treatment of CMF, the surgical options include curettage and excision, with or without filling the bone defect. Curettage alone has resulted in 10%–15% recurrence rate. Wide resection is probably the best method to avoid recurrence, however not all locations allow this procedure [1,7]. CMF is locally aggressive; thus adjuvants such as polymethylmethacrylate are recommended to reduce the rate of recurrence [19].

In conclusion, CMF cannot be differentiated from chondrosarcomas by18F-FDG PET/CT since a high accumulation of 18F-FDG exists in both conditions. Further work with complete investigations of all cases of CMF is needed in order to find standardized diagnostic modalities for the future [20].

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