Osteosarcoma of the Patella Mimicking Giant Cell Tumor: Imaging Features with Histopathological Correlation

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Abstract. Patellar tumors represent an uncommon etiology of anterior knee pain and their diagnosis is often delayed. We present an unusual case of conventional osteosarcoma arising in the patella of a 47-year-old man. The patient presented with a 1-year history of increasing anterior knee pain and swelling. Plain radiographs revealed a multi-locular lytic lesion in the inferolateral side of the patella. Computed tomography scans demonstrated an intraosseous lytic lesion with cortical thinning/breakthrough anteriorly. On magnetic resonance imaging, the lesion exhibited low signal intensity on T1-weighted images and heterogeneous high signal intensity on T2-weighted images. Fluid-fluid levels were also observed on T2-weighted images. Contrast-enhanced fat-suppressed T1-weighted images demonstrated strong enhancement of the lesion. These imaging features were suggestive of a benign condition; however, the diagnosis of osteosarcoma was confirmed by histopathology. After neoadjuvant chemotherapy, the lesion exhibited low signal intensity on T1-weighted images and heterogeneous high signal intensity on T2-weighted images. Fluid-fluid levels were also observed on T2-weighted images. Contrast-enhanced fat-suppressed T1-weighted images demonstrated strong enhancement of the lesion. These imaging features were suggestive of a benign condition; however, the diagnosis of osteosarcoma was confirmed by histopathology. After neoadjuvant chemotherapy, a wide resection with a free anterolateral thigh flap was performed. The patient subsequently underwent adjuvant chemotherapy and had no evidence of local recurrence or distant metastasis six months after surgery. Our case highlights the difficulty in the diagnosis of patellar osteosarcoma and the importance of performing a biopsy before definitive treatment.

Osteosarcoma is the most common primary malignant bone tumor. The patella is an uncommon primary bone tumor site, and patellar tumors are usually benign. Radiographic diagnosis of patellar tumors can often be challenging (1). To our knowledge, there are only 9 case reports of patellar osteosarcoma in the English-language literature (2-8). Herein, we report a rare case of conventional osteosarcoma of the patella in a middle-aged male, with imaging findings that can mimic benign bone tumors, such as giant cell tumor (GCT) and chondroblastoma.

Case Report

A 47-year-old man had complained of a 1-year history of pain and swelling in his left anterior knee region without a trigger episode. About two months later, he visited a local orthopaedic clinic. The anterior knee pain was relieved by conservative management, but the swelling persisted. Three months prior to visiting our hospital, he fell from his bicycle onto his left anterior knee region. He was referred to our hospital because of persistent and progressive pain. Physical examination revealed swelling and tenderness in the inferior side of the left patellar region. The patient could not raise his left lower leg against gravity because of atrophy of the quadriceps femoris. However, we did not observe any skin adhesion, redness, or limitation of the knee joint range of motion. Laboratory data were within normal limits. The patient’s past medical history was unremarkable. Plain radiographs revealed a multi-locular lytic lesion in the inferolateral side of the patella (Figure 1). There was no apparent cortical wall destruction to the patellofemoral joint. Computed tomographic (CT) scans demonstrated an intraosseous lytic lesion with cortical thinning/breakthrough anteriorly (Figure 2). On magnetic resonance imaging (MRI), the lesion exhibited low signal intensity on T1-weighted images (Figure 3A) and a heterogeneous high signal intensity on T2-weighted images (Figure 3B). The margins of the lesion were relatively well-defined, but anterior soft tissue extension was suspected. Fluid-fluid levels and thin septations were also...
observed on T2-weighted images. Contrast-enhanced fat-suppressed T1-weighted images demonstrated strong enhancement of the lesion (Figure 3C). Tc-99m Hydroxymethylenediphosphonate bone scintigraphy showed no abnormal uptake except for the left patella (Figure 4). Based on clinical and radiological features, a benign bone tumor was suggested, including GCT with secondary aneurysmal bone cyst (ABC) and chondroblastoma with secondary ABC.

Unexpectedly, the rapid intraoperative pathological diagnosis was a malignant tumor; therefore, open biopsy was performed and the extended operation was canceled. Microscopic examination of the open biopsy revealed that the tumor demonstrated diffuse proliferation of atypical mesenchymal cells, with large, hyperchromatic nuclei and prominent nucleoli. Irregular osteoid formation (Figure 5A), pleomorphic malignant fibrous histiocytoma (MFH)-like areas (Figure 5B), and telangiectatic areas (Figure 6) were also found. Mitotic figures, including abnormal forms, were frequently encountered. The histopathological features were indicative of conventional osteosarcoma with areas of MFH-like and telangiectatic components.

No evidence of distant metastasis was found on integrated positron-emission tomography/CT scan. The maximum
The standardized uptake value of the left patellar lesion was 3.56. The patient received neoadjuvant chemotherapy with high-dose methotrexate, cisplatin, and doxorubicin. The follow-up MRI after neoadjuvant chemotherapy showed a reduced size of the lesion. Because the tumor did not spread into the knee joint on MRI, we performed a wide resection with a free anterolateral thigh flap, including the quadriceps tendon, patella, medial and lateral patellar reticulum, patellar tendon, and intrapatellar fat pad. No tumor invasion of the knee joint was observed. All resected surgical specimens were sampled, fixed in formalin and embedded in paraffin, then examined microscopically. We observed areas of extensive fibrosis with small vessel proliferation. Less than 10% of the sample consisted of viable tumor cells. Three weeks postoperatively, adjuvant chemotherapy was performed for a total of two cycles according to the Neoadjuvant Chemotherapy for Osteosarcoma (NECO)-95J protocol. The patient had no evidence of local recurrence or distant metastasis six months after surgery.

**Discussion**

Patellar tumors are an uncommon cause of anterior knee pain and their diagnosis is often delayed (9). In a review by Ferguson et al., eight cases of primary tumors of the patella were identified in a consecutive series of 587 patients who were surgically-treated for benign or malignant bone tumor (4). Six cases were benign (five GCTs and one chondroblastoma), and there were only two malignant tumors (one osteosarcoma and one MFH). Recently, Casadei et al. also reported that the most frequent benign patellar tumor is GCT, followed by chondroblastoma (1). On the other hand, malignant tumors are much less common than benign lesions,
and metastases are the most frequent malignant tumors of the patella (1). Primary malignant lesions include mostly osteosarcoma, hemangioendothelioma, and lymphoma (1).

To the best of our knowledge, nine cases of osteosarcoma in the patella have been described in the English-language literature (2-8). Compared to osteosarcomas occurring elsewhere in the skeleton, the age of such patients are relatively high. The peak incidence is in the fourth or sixth decade of life. Chida et al. suggested that this age distribution may be caused by pre-existing conditions such as genetic disorder and irradiation (2). However, the presence of pre-existing conditions was not found in our patient. Pain and swelling in the front of the knee are the most frequent presenting symptoms, as in our case. Radiographically, osteosarcomas often involve the majority of the patella and margins of the lesion are ill-defined (2, 8). An aggressive periosteal reaction is absent in the patella. On MRI, fluid-fluid levels have not been reported in conventional osteosarcomas of the patella (1), except in our case.

The histopathological diagnosis of the current case was conventional osteosarcoma with telangiectatic and MFH-like components. One of the reasons it was difficult to make a preoperative diagnosis seemed to be related to the presence of telangiectatic osteosarcoma components, which can be radiologically confused with ABC. Pure telangiectatic osteosarcoma is a rare subtype, accounting for fewer than 4% of all osteosarcomas. On T2-weighted images, telangiectatic osteosarcomas show several cystic foci, fluid levels, and an extraskeletal extension of the tumor (10). Only one case of telangiectatic osteosarcoma of the patella has been reported.
(8). The reported case presented as a large lesion arising from the patella with smaller lesions of the tibia and femur, as well as pulmonary metastasis.

The differential diagnosis of the current case includes GCT and chondroblastoma. GCT typically occurs in the third or fourth decade of life. Radiographic features in the patella are similar to other sites of the skeleton (11). Radiographically, GCTs usually demonstrate eccentric, purely lytic lesions lacking internal mineralization. The cortex is frequently destroyed, and the lesion expands to the soft tissue (12). GCTs often involve more than 75% of the patella (11). Chondroblastoma typically occurs in the second decade of life, with a male predominance. Radiographic appearances in the patella do not significantly differ as compared to other skeletal locations (11). Radiographically, chondroblastomas usually demonstrate well-defined, round or oval, lytic lesions with a surrounding thin sclerotic rim. Internal mineralization may sometimes be present. A periosteal reaction is absent in the patella (1). Both GCT and chondroblastoma can demonstrate fluid-fluid levels on MRI due to secondary ABC formation (12). In the current case, it was impossible to distinguish these conditions from osteosarcoma on imaging alone. These findings suggest that a biopsy is necessary to obtain a definite diagnosis before treatment.

Recently, much progress has been made in the application of combination chemotherapy for osteosarcoma. Multi-agent chemotherapy for high-grade osteosarcoma has had a dramatic impact on outcome (13, 14). In the pre-chemotherapy era, 80% of patients solely treated with surgery died of disease, whereas 70% of current patients become long-term survivors (13-15). Localized distal disease, >90% chemotheray-induced tumor necrosis, and complete resection are positive prognostic factors associated with a 5-year survival rates of >80%. Previous reports of patellar osteosarcoma cases describe poor prognosis due to distant metastasis and chemoresistance (2, 5, 6, 8). Our case responded well to combination neoadjuvant chemotherapy. Fewer than 10 % of cells were viable in the extended-resection surgical specimen. No apparent systemic metastasis was found during preoperative whole-body scanning, suggesting good prognosis for the present patient.

In summary, we have described a unique case of conventional osteosarcoma of the patella mimicking GCT with secondary ABC in a middle-aged patient. Physicians should be aware of the possibility of osteosarcoma when confronted with a patellar tumor presenting with fluid-fluid levels on MRI.

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References


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