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Case Report

Primary bone lymphoma of patella: A case report and review of literature

Jami Jadidi, MD[a,*], *Fardad Behzadi, BSc*[b], *Maziar Sighary, MD*[a], *Mohamed Alshal, MD*[c], *Srinivas Kolla, MD*[a], *Scott A. Lehto, MD*[a]

[a] *Department of Radiology, SUNY Downstate Health Sciences University, 450 Clarkson Ave, Brooklyn, NY 11203, USA*

[b] *Department of Radiology, Brigham Women's Hospital, 75 Francis St, Boston, MA 02115, USA*

[c] *Department of Pathology, SUNY Downstate Health Sciences University, 450 Clarkson Ave, Brooklyn, NY 11203, USA*

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ABSTRACT

Primary bone lymphoma (PBL) is rare bone disease that accounts for very small number of all primary bone tumors. Among the described sites of PBLs, the patella is an extremely rare example. To date, only a few cases of PBL affecting the patella have been reported. Clinically, these tumors have very similar presentation of pain, decreased range of motion and swelling and, sometimes, pathologic fractures. On radiographs, skeletal lymphoma commonly manifests as osteolytic lesions with ill-defined margins affecting the metaphysis of axial long bones. We present a rare case of patellar adult T-cell lymphoma/leukemia in a 58-year-old female who presented with left-knee pain and swelling. Computed tomography and magnetic resonance imaging revealed diffuse marrow replacement by a lesion with aggressive features. PET scan demonstrated neoplastic range hypermetabolic FDG uptake within this lesion. Ultrasound-guided biopsy was consistent with PBL.

Introduction

Primary bone lymphoma (PBL) was first described in 1928 as a type of lymphoma with malignant lesions affecting bones as primary site [1]. PBL is a rare bone disease that accounts for only 2% of all primary bone tumors [2]. This number in pediatric population is 3%-9% of non-Hodgkin lymphomas which could be attributed to the rapid growth rate in this patient population [3,4]. There seems to be a male dominance in the number of recognized cases; males are twice more likely to get diagnosed with PBL as opposed to females [16]. Most of the PBL cases manifest themselves clinically at age >30 years [16]. Among the reported cases, non-Hodgkin diffuse large B-cell lymphoma comprises the majority [5]. The patella is an extremely rare primary site for formation of PBL. To date, only 4 cases of PBL involving the patella have been reported among the adult population, with at least one case being reported

* Corresponding author.
E-mail address: jami.jadidi@downstate.edu (J. Jadidi).

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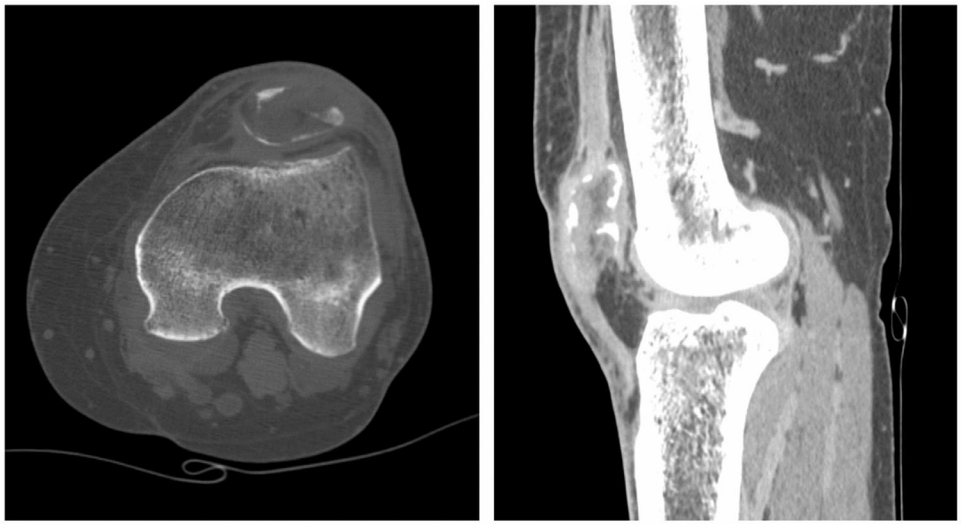

Fig. 1 – Noncontrast CT of the left patella shows a lytic lesion both on axial and sagittal views. Low-attenuated marrow replacing lesion of almost the entire patella with significant cortical loss is evident. Focal area of soft tissue extension to the patellar tendon is also noted.

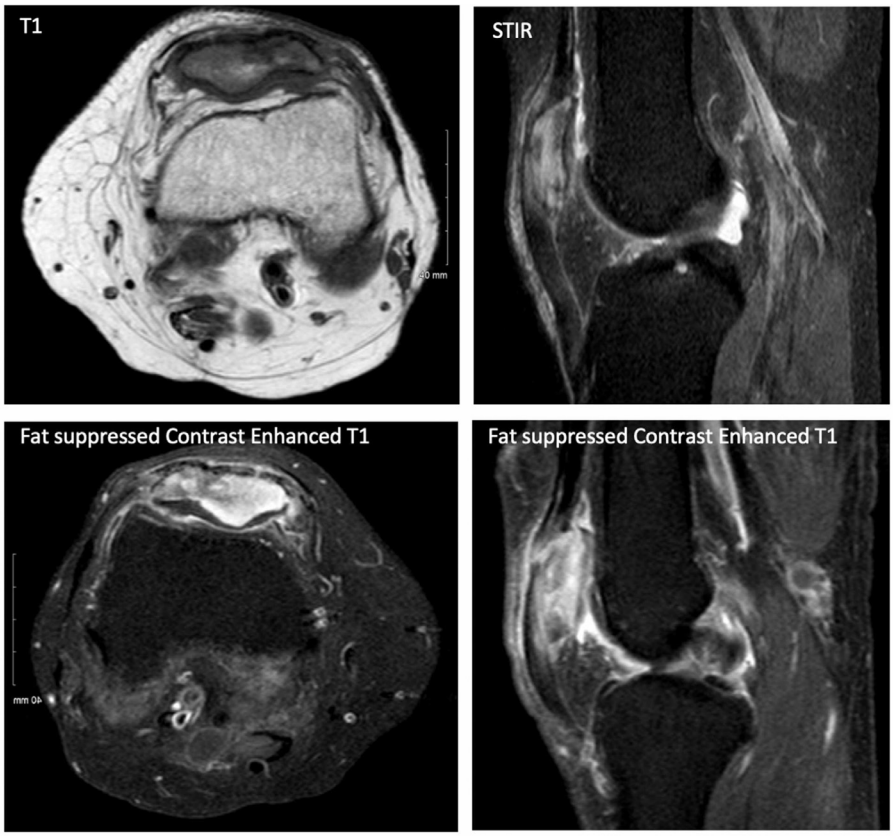

Fig. 2 – MRI of the left patella revealing marrow replacement of the patella by an aggressive lesion featuring decreased T1 signal, increased STIR signal and avid postcontrast enhancement. There is cortical disruption anteriorly with surrounding soft tissue edema and enhancement of distal quadriceps and proximal patellar tendons.

among the pediatric population [6,7,11,12]. Radiologic manifestations of PBL share common features with those of histiocytic lymphoma, including osteolytic lesions with ill-defined margins [13].

We report a case of a 58-year-old female who initially presented with chronic left-knee pain. Imaging features and histological correlation were most consistent with PBL, specifically, adult T-cell lymphoma/leukemia of the patella.

Case report

A 58-year-old woman with no significant history presented with left-knee pain and limited range of motion for a 5-month period. Physical examination confirmed limited range of motion; no other significant finding was observed. Radiograph imaging workup corresponded with lytic lesion of the patella. Computed tomography demonstrated a lytic lesion with ill defining borders, raising suspicion of malignancy (Fig. 1). Magnetic resonance imaging revealed marrow replacement by a lesion demonstrating decreased signal on T1-weighted images, increased signal on STIR and avid contrast enhancement (Fig. 2). Cortical disruption with surrounding soft-tissue edema was also noted. Subsequently, patient underwent an ultrasound guided core-needle biopsy of the patellar lesion. Histopathological studies demonstrated the presence of an atypical lymphocyte proliferation involving bone in a background of fibrosis and adipocytes. The atypical lymphocytes were highly pleomorphic with hyperchromatic nuclei. Immunohistochemical analysis of these atypical lymphocytes demonstrated positive expression for CD45, CD2, CD3, CD4, CD5, CD8(variable), CD25, FOXP3 (minor subset), and with partial loss of CD7. Ki-67 proliferation index was high (60%-70%) (Fig. 3). Enzyme Immunoassay of the patient's serum detected Human T-cell Lymphotropic Virus-I/II (HTLV I/II) antibodies and Western Blot confirmed the presence of HTLV-I infection. Bone marrow biopsy revealed a normocellular bone marrow with no evidence of T-cell lymphoma. Overall, these findings were most consistent with the diagnosis of primary bone adult T-cell lymphoma/leukemia of the patella. Finally, a whole-body PET scan follow-up revealed neoplastic range hypermetabolic FDG uptake within the left patella. A single hypermetabolic node within the popliteal fossa, representing regional lymph node metastasis, was also observed (Fig. 4). Nonspecific sub-centimeter left inguinal node demonstrating low level FDG uptake, likely physiologic or inflammatory in etiology was also noted. Overall, these findings were most consistent with PBL involving the left patella. Subsequently, our patient was planned for intensive chemotherapy regimen followed by radiation to the left patella. She was also scheduled for lumbar

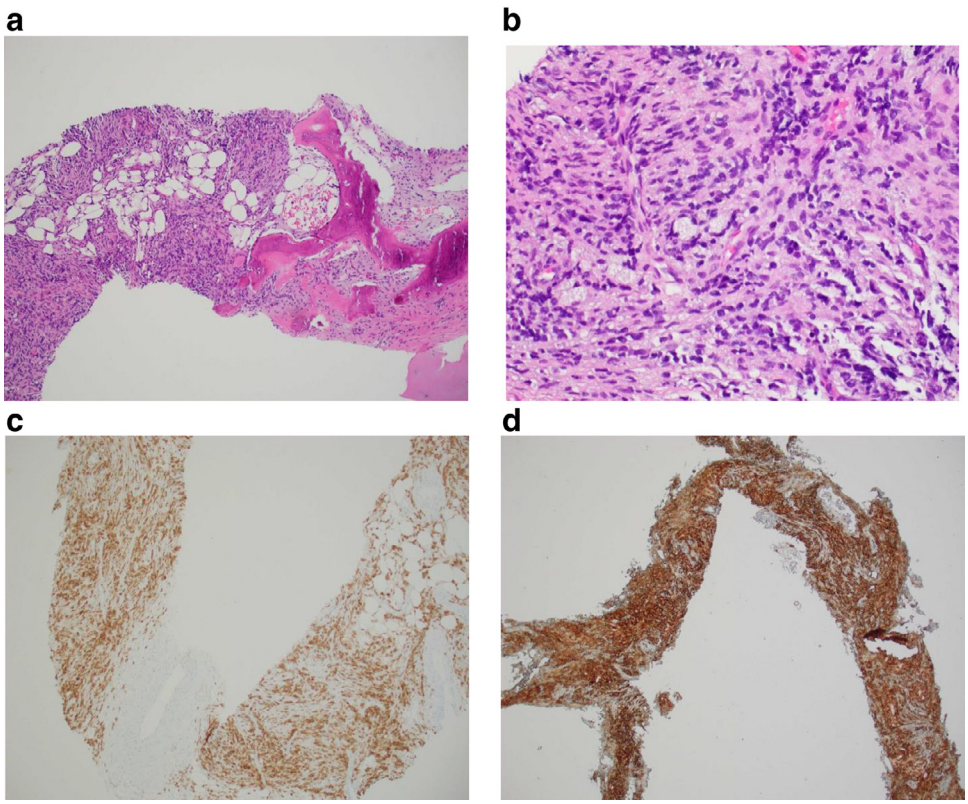

Fig. 3 – Morphological features of the primary bone lymphoma. (a) Atypical lymphocyte proliferation involving the bone in a background of fibrosis and adipocytes (H&E, 100x). (b) The infiltrating atypical lymphocytes are highly pleomorphic with hyperchromatic nuclei (H&E, 400x). Immunohistochemical stains show the atypical lymphocytes are positive for CD3 ((b), x100) and CD4 ((c), H&E, x100), supporting the diagnosis of T-cell lymphoma.

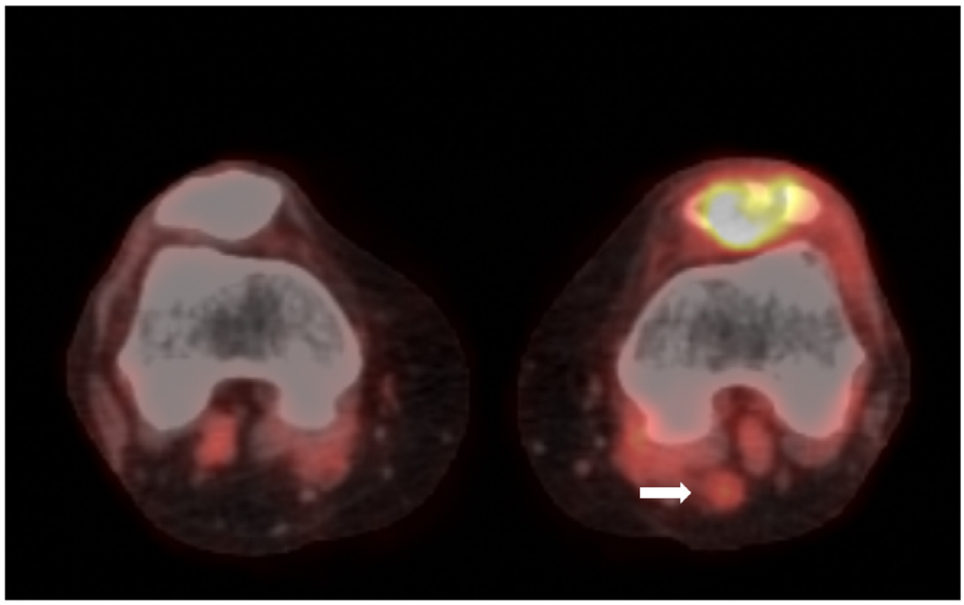

Fig. 4 – CT-PET demonstrating a patellar lesion with hypermetabolic FDG uptake to maximal SUV of 10.6. Also noted is an 8 mm soft tissue density within the popliteal fossa with increased FDG uptake to 2.6 SUV representing lymph node involvement (white arrow).

puncture (LP) to assess for central nervous system (CNS) disease with consideration for intrathecal chemotherapy.

Discussion

Primary tumors of patella are generally rare among bone tumors, forming about 0.12% of all bone tumors, and mainly have benign etiology [8,9]. It has been estimated that 73% of patella tumors are benign while 27% have been reported to be malignant [8]. Among the benign tumors, giant cell tumor and chondroblastoma compose the vast majority of tumors [8]. Giant cell tumors are estimated to compose about 33% of all patellar tumors, while Chondroblastoma's compose roughly about 16% of all patellar tumors [8]. Patellar metastases are even rarer compared to primary tumors of patella [10]. A review of the reported cases from 1960 to 2016 has only resulted in 44 patellar metasets cases [10]. Of these metastases cases bone metastases disease originating from the lungs have been reported to be the leading cause with 17 reported cases [10].

PBL is defined as malignant bone tumors that affect a wide range of bones without affecting other internal organs or distant lymph nodes [14]. To this date, only 2 cases of PBL with concurrent magnetic resonance imaging have been published [12,15]. Our case meets the diagnostic criteria defined by Dosoretz: PBL of left patella with only one regional lymph node being affected without internal organ involvement [14].

PBL lesions generally manifest with localized pain, swelling, and, in some cases, decreased range of motion [15]. Radiographically, PBLs reveal lytic bone lesions [15]; this is consistent with the primary patellar computed tomography finding in our case.

Prognosis of PBL is generally promising, with a 5-year survival rate of 58%-74% in adult population [16]. In another study published in Japan, the 3-year survival rate was even higher, at 63%-84% [17]. Historically, the treatment of choice for PBL has been radiation and chemotherapy [18]. Patellectomy can also be performed in cases with isolated lesions. However, it is preferred to try noninvasive methods such as chemotherapy and radiotherapy first, since both have had good outcomes, as reported in the literature.

In conclusion, we diagnosed a rare case of PBL, particularly a subtype of T-cell lymphoma of the patella. With increasing numbers of reported PBL, it is important that PBL is included in possible differential diagnoses when encountering lesions with those described characteristics found in our case.

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