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Case Report

Parosteal osteosarcoma with focal fatty metaplasia: A case report

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ABSTRACT

Parosteal osteosarcoma originates on the surface of long bones. The juxtacortical variety is one of the most common ones and accounts for about 5% of all osteosarcomas. We report the case of a 34-year-old female patient with a rare variant of parosteal osteosarcoma. Because of the less aggressive biological behavior, it is important to know it, in order to recognize and differentiate it from benign osseous lesions.

Introduction

Parosteal osteosarcoma (PO) is a typically well-differentiated subtype of osteosarcoma, with slow growth and low grade malignancy that do not tend to metastasize. PO is the most common type of osteosarcoma arising on the bone periosteum and accounts for 2%-4% of all primary malignant bone tumors, approximately 4%-6% of all osteosarcomas, and 70% of surface osteosarcomas [1]. PO mainly affects adults between the ages of 20-30 years and exhibits a slightly higher prevalence in women. The most common sites affected by PO include the knee, accounting for about 70% of cases [2], and specifically the posterior aspect of the distal femur, followed by the proximal tibia, proximal humerus, fibula, radius, and ulna; only 6% of all POs are observed in the skull, the spine, and pelvis [3].

The radiographic feature of osteosarcoma includes a lobulated exophytic mass with central dense ossification adjacent to the bone usually in the posterior aspect of the distal femur, sparing the medullary canal. Computed tomography scans

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reveals well-defined, hyperdense bony mass with patchy radiopacification. Sometimes PO shows highly differentiated areas of fatty tissue within the marrow and uniform bony structure, making difficult the diagnosis.

PO originates from the outer fibrous layer of the periosteum and is mainly composed of fibroblasts.

Grossly, the tumor appears as a hard lobulated mass attached to the underlying external layer of periosteum, and may contain nodules of cartilage within the substance of the tumor or an incomplete cartilage cap at the surface [4]. Microscopically, PO is characterized by hyalinized fibrous stroma with a low cell content without substantial nucleus polymorphism and variably dense bony trabeculae [3]. In some cases, the diagnosis of PO can be complicated by the presence of highly differentiated areas with fatty tissue within the marrow and uniform bony structure [3].

Here, we report a case of focal fatty metaplasia identified within a PO lesion. Given a lack of previous reports on this rare condition, we offer a characterization of this rare histomorphological variation to enrich the morphological spectrum of PO.

Case description

A healthy 34-year-old woman was referred to our orthopedic oncology clinic for a mass that was identified in her right tibia on radiography after sudden injury of her right lower limb 2 months prior. The patient did not experience any pain or swelling related to the mass. A biopsy performed at another hospital did not reveal any histologic evidence of malignancy.

On physical examination, the patient presented with a hard, poorly circumscribed, painless, and immobile palpable mass at the posterior aspect of the proximal right tibia. The area surrounding the mass was slightly warm, but no changes in the skin were noted. The patient had normal range of motion of the right knee and no abnormal findings on hematologic or biochemical studies.

Radiography of the tibia revealed a well-circumscribed bone-forming lesion associated with the external periosteum of the proximal tibia (Fig. 1). Computed tomography showed no medullary involvement and confirmed that the bony mass arose from the periosteum, exhibiting a lower density than the tibial cortex. The tibial cortex appeared to be intact. All findings were compatible with PO. Additionally, 2 areas of fatty density were observed in the osteoid tumor (Fig. 2).

The patient subsequently underwent resection of the affected segment of the proximal right tibia (19 cm below the tibial plateau) and reconstruction of the right knee and tibia by prosthesis implantation. The hematoxylin and eosin stained histologic diagnosis was PO with focal fatty metaplasia (Fig. 3). Good histologic margins were achieved and there was no tumor recurrence or metastasis after the resection.

Okada et al [5] defined criteria for the radiological diagnosis of PO to include lesions arising from the surface of the bone with good differentiation of the tumor on histology (grade 1 or 2); a well-formed osteoid within a spindle-cell stroma; and, in cases of medullary involvement, occupation of

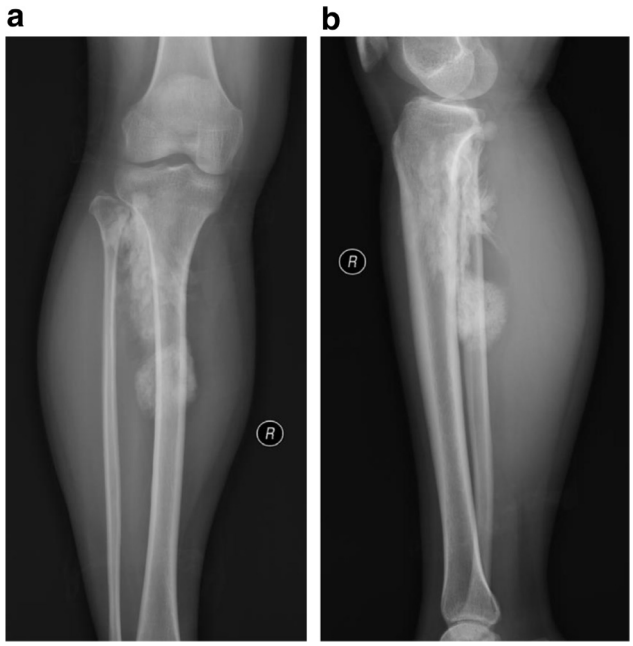

Fig. 1 – Antero-posterior (a) and lateral (b) radiography of the right tibia showing an ossifying mass surrounding the posterior and lateral aspects of the upper tibia. Due to overlying aspect of tumor mass, an involvement of the bone trabeculae cannot be ruled out on the basis of X-ray radiography solely.

<25% of the medullary cavity. A thin radiolucent zone is seen between the tumor mass and host bone cortex in >50% cases, and the cortex of the host bone can be normal, thickened, or destroyed.

In some cases of PO, radiolucent areas are noted within the tumor, presumably indicating areas of dedifferentiation. Bertoni et al [6] studied the significance of radiolucencies in PO and hypothesized that a majority of peripheral lucent areas were comprised of low-grade malignant cartilaginous or fibrous tissue mixed with fat and bone trabeculae, whereas deeper radiolucencies corresponded to areas of high-grade dedifferentiation.

Cytogenetic analysis of PO has demonstrated supernumerary ring chromosomes containing gain of 12q13-15 sequence. This characteristic cytogenetic abnormality is uncommon in conventional osteosarcoma. Such ring chromosomes are noted in other low-grade malignant mesenchymal neoplasms such as well-differentiated liposarcoma and dermatofibrosarcoma protuberans [7].

SAS gene is located in q13-15 region of chromosome 12 and is found to be amplified in surface osteosarcomas, also in the parosteal subtype [8]. Other genes, frequently coamplified or over expressed in parosteal osteosarcoma include CDK4 and MDM2 [9].

The differential diagnosis of PO usually includes some benign conditions such as osteochondroma and myositis ossificans; the first usually presents corticomedullary continuity with the underlying medullary canal, while myositis

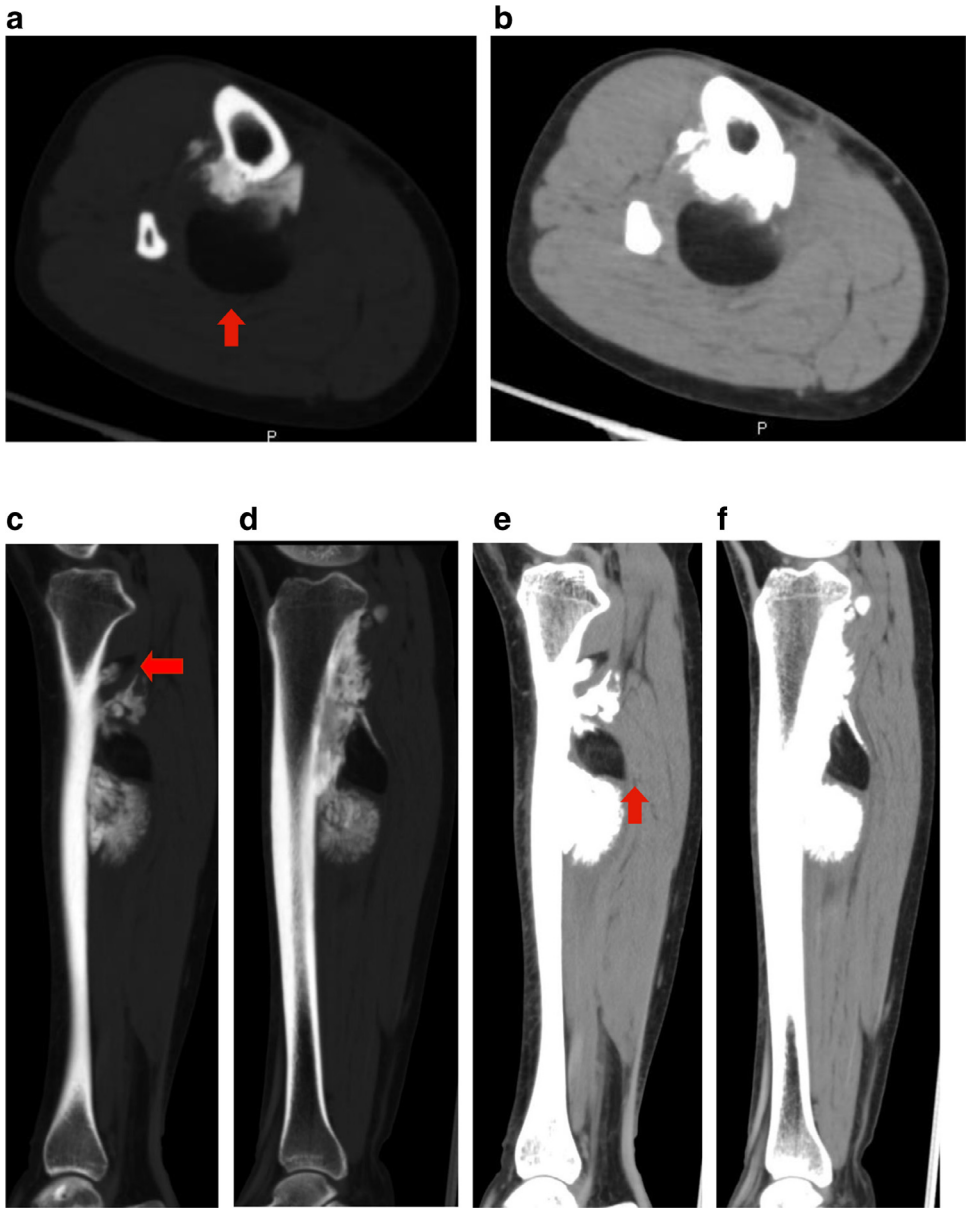

Fig. 2 – Axial (a, b) computed tomography and sagittal reconstruction images (c-f) showing an ossifying mass arising from the periosteum with no spongious bone infiltration.
Note the 2 areas of fatty density within the tumor.

ossificans presents a peripheral ossification, inverse of the PO ossification pattern, with calcifications centrally located [10].

Diagnosing a PO needs a high index of suspicion. All clinicoradiologically doubtful lesion needs to be biopsied to confirm the diagnosis.

The natural history of PO is relatively benign. Its optimal treatment is wide surgical resection, with an overall survival rate of 80%-90%. Incomplete removal is associated with local recurrence and increased risk of metastasis [5].

In the present case, we observed radiolucent areas in the tumor that were later identified on histology as focal fatty metaplasia. Because of the less aggressive biological behavior, it is important to know this variant of parosteal osteosarcoma, in order to recognize and differentiate it from benign osseous lesions.

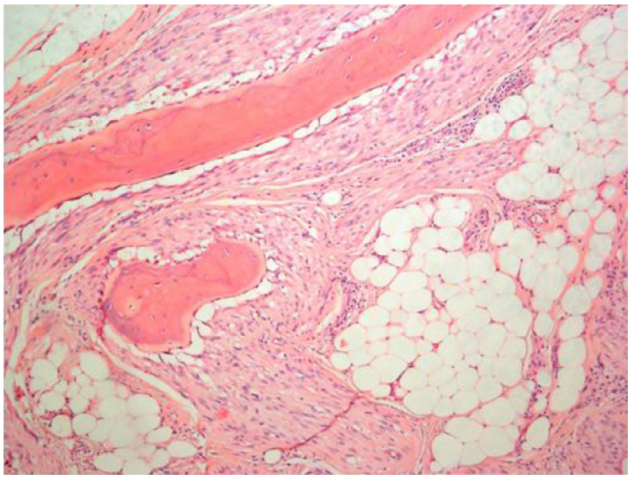

Fig. 3 – Sheets of metaplastic adipocytes among the spindle tumor cells of a parosteal osteosarcoma (H&E stain, ×100).

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