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Case Report

Bizarre parosteal osteochondromatous proliferation of the femur: A case report

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ABSTRACT

A 54-year-old man initially presented with left distal thigh pain during walking. Imaging analysis revealed a diffuse calcified or ossified mass adjacent to the medial cortex of the distal femur and absence of continuity with the medulla. We performed resection biopsy. Histological examination revealed a large amount of hypercellular cartilage showing transformation to trabecular bone and BPOP was diagnosed. Postoperative course was uneventful and he remained free of recurrence. The method of resection should depend on the stage of reactive proliferation and whether the lesion is pathologically immature or mature. We also provide a brief review of the literature.

1. Introduction

1.1. Case report

A healthy 54-year-old Japanese man presented with left distal thigh pain during walking 3 months prior to surgery. His past history was unremarkable and he reported no trauma in the thigh. Clinical examination revealed a tender and non-mobile hard mass measuring approximately 3 cm in diameter. Plain radiographs and computed tomography revealed a diffuse calcified or ossified mass adjacent to the medial cortex of the distal femur and absence of continuity with the medulla (Fig. 1). On magnetic resonance imaging, the lesion demonstrated a low signal intensity on T1-weighted images and a high signal intensity with a low signal intensity on the surface on T2-weighted images (Fig. 2). Therefore, our differential diagnosis included osteochondroma, bizarre parosteal osteochondromatous proliferation (BPOP), parosteal osteosarcoma, and parosteal chondrosarcoma. We performed resection biopsy to obtain a definite pathological diagnosis. The lesion was elastic, hard, and nonmobile, measuring 3 × 3 cm and consisting of cartilage-like tissue (Fig. 3). Histological examination revealed a large amount of hypercellular cartilage showing transformation to trabecular bone, with several spindle cells in the intertrabecular spaces. There was also an area composed of a mixture of bone, cartilage, and fibrous granulation tissue (Fig. 4). Therefore, the possibility of malignant tumors was ruled out, and a diagnosis of BPOP was established. One year and three months after surgery, the patient has had no subsequent pain and is free of local recurrence.

2. Discussion

Whether BPOP is a neoplasm or a reactive proliferative lesion is unclear.[1–3] Dhondt et al described a spectrum of reactive lesions with differences being attributable to the degree of maturation: florid reactive periostitis (FRP), then BPOP, and finally turret exostosis.[1] Yuen et al claimed that the recurrence rate depends on factors related to breaching of the stage of evolution and maturation of the process.[1,4,17] Additionally, Dhondt et al retrospectively analyzed the radiographical appearance of BPOP and demonstrated that its lesions demonstrate a spectrum of reactive changes that include FRP and turret exostosis.[1] On the other hand, BPOP has also been considered to be a neoplasm owing to the high recurrence rate; moreover, several studies have reported chromosomal aberrations such as t(1;17)(q32;q21), t(1;17)(q42;q23) translocation.[2,5]

BPOP can be easily misdiagnosed as osteochondroma, FRP, myositis ossificans, parosteal chondrosarcoma, or parosteal osteosarcoma on the basis of its appearance on preoperative imaging and pathological examination.[6] Moreover, osteochondroma is the most common benign tumor of the skeletal bones, and they are more likely to be found in the metaphyseal region of long bones.[5,7,8,13] On imaging analysis, BPOP lesions show a lack of continuity with the medulla on computed

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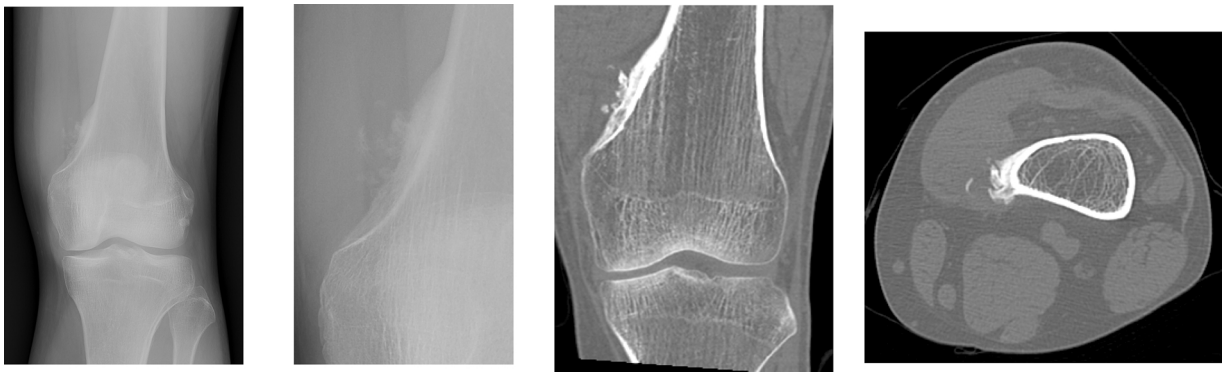

Fig. 1. X-ray and CT revealed diffuse calcified or ossified mass adjacent to the medial cortex of the distal femur and absence of continuity with the medulla.

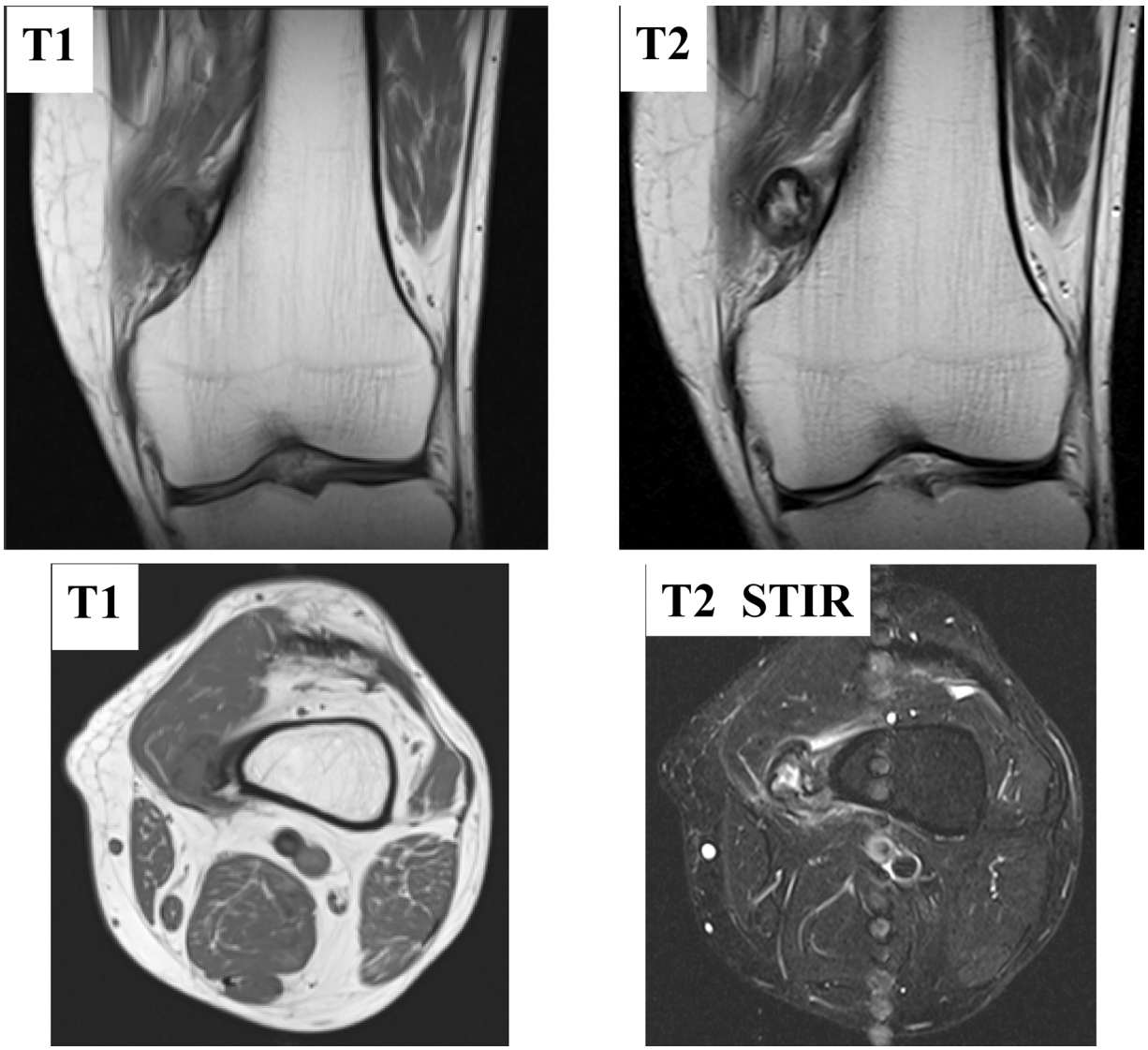

Fig. 2. MRI revealed the lesion had a low signal intensity on T1-weighted images, and a high signal intensity with a low signal intensity on the surface of the lesion on T2-weighted images.

tomography and magnetic resonance imaging, while osteochondroma maintains a continuity.[6,7,12] This is considered to be the characteristic difference between the two conditions. Because the lesion observed in the present case had no clear continuity with the medulla, along with its radiographic appearance, BPOP was considered.

On pathological examination, BPOP demonstrates cartilage at the margins of the lesion and irregular mature bone or evidence of ossification just beneath the trabecular bone.[3,6,12] The marginal cartilage resembles reactive fibrocartilage.[5] Because the cartilaginous tissue in BPOP derives from parosteal mesenchyme, marrow discontinuity between the BPOP and the original bone can be seen. Trabecular bone shows an irregular distribution and represents incomplete