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

Fig. 3. The lesion was consisted by a cartilage like tissue and its interior was composed of osteoid tissue in continuity with the cortical bone. The lesion was elastic, hard, nonmobile mass measuring 3 × 3 cm.

Fig. 4. Histological examination revealed a large amount of hypercellular cartilage showing transformation to trabecular bone, with frequent spindle cells in the intertrabecular spaces. There was also an area composed of a mixture of bone, cartilage, and fibrous granulation tissue.

endochondral ossification. Some of the chondrocytes exhibit mild atypia, such as nuclear enlargement and binucleation. In this study, the imaging findings and pathological findings correspond to those of previous reports. On the other hand, osteochondroma shows complete endochondral ossification with overgrowth of hypertrophic chondrocytes. The marginal cartilage is the hyaline cartilage and it does not exhibit cytologic atypia.⁵ The hypertrophic chondrocytes observed in osteochondroma produce trabecular bone that subsequently connects with the original bone marrow. However, in general, the trabecular bone growth in BPOP is more active and extensive than that of osteochondroma. BPOP has multiple cartilaginous tissues that are immature and diffusely scattered, while osteochondroma has a thick cartilage cap. These findings suggest that BPOP may be associated with higher a recurrence rate than osteochondroma.⁹

In the English literature, only 6 cases of BPOP arising in the femur have been reported thus far, to the best of our knowledge,^{10,11,3,6,12} Although rare, it is important to consider the possibility of BPOP occurring in the femur. Although the rate of recurrence after primary excision is reported to be about 50% and that after the first recurrence about 20%,^{5,6,12} no study has investigated or reported the adequate method of resection for BPOP. In general, if the condition is asymptomatic, observation alone is sufficient.⁵ However, almost all cases of BPOP are symptomatic and demonstrate pain, localized swelling, and impaired function. Several studies have reported various surgical excision approaches.^{3,5-7,9,13-17} First, simple excision (marginal excision) is widely known.^{3,6,12} It is indicated for symptomatic BPOP cases and remains the preferred treatment for primary lesions. However, simple excision is known to be associated with a high rate of recurrence

(20–55%) after surgery.^{5,6,12} The second treatment option is resection of the capsule of the lesion and decortication of the underlying cortical bone. This approach involves a much lower recurrence rate, about 10%. Joseph et al reported that this method did not result in subsequent recurrence in a patient initially treated with simple excision.³ The third treatment option is elective surgery. Dhondt et al reported that radiographical symptoms of BPOP can be observed only after 6 months; moreover, the lesion demonstrates a spectrum of reactive changes from FRP to turreted exostosis.¹ They also reported that the period of observation until actually deciding to perform the second surgery is more than 6 months. Additionally, the Japanese literature reports an elective time of over 6 months and identification of lesion maturation on radiography on postoperative pathological examination. Gursel et al described that aggressive wide local resection and/or ray amputation may be required to limit lesion progression and to restore function in cases of remarkable bone destruction, swelling, and impaired function.⁷

In the present case, the patient experienced no subsequent pain and is free from local recurrence 1 year and 3 months after the surgery. Because the lesion was mature, on the basis of the pathological findings, resection may have been the correct approach. It has reported that surgical excision with wide margins is adequate because of the high recurrence rates,¹² however, the findings of the present case suggest that the method of resection should depend on the stage of reactive proliferation (from FRP to turreted exostosis) and identifying whether the lesion is pathologically immature or mature.

3. Conclusion

We presented a rare case of BPOP of the femur. Although rare, it is important to acknowledge that BPOP can occur in the femur. Previous reports have described a high rate of local recurrence following surgical excision; however, our case did not demonstrate local recurrence after more than a year. Nonetheless, careful follow-up is essential.

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Conflicts of Interest

The authors have none to declare.

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