



Bizarre Parosteal Osteochondromatous Proliferation: Report of a Case with Hallmark Characteristics

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Abstract

Introduction: Bizarre parosteal osteochondromatous proliferation (BPOP), also known as the Nora's lesion, is a part of the spectrum of reactive lesions with a difficult diagnosis. To date, only limited number of Nora's lesions have been reported in the literature. Here, we report a case of Nora's lesion and discuss the differential diagnosis of the case.

Case Presentation: A 34-year-old male that was referred to the hand clinic of our center with a painful lump at the dorsoulnar aspect of the first metacarpal bone of his left hand. The diagnosis of BPOP was suspected using its clinical and radiographic characteristics. Subsequently, excisional biopsy was performed and the extracted lesion was sent to the pathology for definitive diagnosis. The histopathologic evaluation confirmed the diagnosis of BPOP. One year follow-up of the patient showed no radiographic or clinical sign of recurrence.

Conclusions: BPOP can be confused with malignant lesions such as parosteal osteosarcoma and chondrosarcoma. Thus care should be taken to combine the radiographic and pathologic information in the correct diagnosis of this lesion, especially when the BPOP presents with atypical features.

Keywords: Bizarre Parosteal Osteochondromatous Proliferation, Differential Diagnosis, Metacarpal Bone, Hand

1. Introduction

Nora's lesion or bizarre parosteal osteochondromatous proliferation (BPOP) is a non-neoplastic lesion presented as the outgrowth from the cortical surface. First described by Nora et al. in 1983 (1), the mass is a fibro-osseous proliferation with a cartilaginous cap that affects patients of any age with no sex predilection. Due to its rapid growth and bizarre radiologic/histologic appearance, the differential diagnosis of BPOP is wide and mainly includes soft-tissue chondroma, osteochondroma, parosteal/periosteal osteosarcoma, and periosteal chondrosarcoma (2). Their atypical histologic appearance and the high rate of recurrence make further complicated their differential diagnosis from malignant lesions (3). Although periosteal trauma or ischemia is suggested as the etiologic factor for BPOP, numerous questions regarding its etiology, diagnosis, and treatment remain unresolved (2). Case reports of BPOP are the available approach to unwrap the uncertainties about BPOP. In addition, they raise awareness regarding the correct diagnosis and management of this lesion.

Here, we report a case with BPOP in a 34-year-old male with a 3-year-history of pain at the first metacarpal bone of his left hand. We also discuss the histologic, radiologic, and clinical features of the case, as well as the differential diagnosis, management, and outcome.

2. Case Presentation

A 34-year-old male referred to the hand clinic of our center with a mass on the 1st metacarpal bone of his left hand and 3-month-history of pain exacerbated following the activities. The patient remembered no antecedent trauma. In physical examination, a tender lump with palpation was present over the ulnar side of the metacarpal bone that was firm and immobile. Neurovascular examinations did not reveal any abnormality. Thumb's range of motion was normal as well.

In the plain radiograph (Figure 1) and computed tomography (CT) scan (Figure 2), a parosteal flame like bony proliferation and calcification was evident, extending from the proximal side of the ulna to the first metacarpal

bone. The soft-tissue component of the mass was evaluated by magnetic resonance imaging (MRI) (Figure 3). A nodular soft-tissue swelling was detected around the bony proliferation and calcification. Based on the clinical and radiologic findings, the diagnosis of BPOP was suspected and the mass was surgically excised.



Figure 1. Anteroposterior and lateral radiographs of the left thumb demonstrating the BPOP lesion

The resected mass was sent to the pathology department for histopathologic evaluations. Microscopic evaluations revealed deposition of hyper-cellular woven bone in a fibrous context in addition to the diffuse proliferation of monomorphic spindle cells with small nuclei and pale chromatin pattern. Minimal cartilage formation was surrounded by more mature areas with the trabecular arrangement of bone (Figure 4).

The cumulative pathologic findings confirmed the diagnosis of BPOP. During the one year post-operative follow-up of the patient, no complaint was reported by the patient and no recurrence was observed in the follow-up radiographs.

3. Discussion

Some authors believe that BPOP represents a neoplastic lesion rather than a reactive lesion (2). This conception is supported with the identification of an abnormal karyotype (4) and non-clonal abnormalities of chromosomes 2,

8, and 14 in some lesions (5). Whatever the etiology of BPOP is, radiology alone is reported to be sufficient for the diagnosis of typical BPOP. However, the diagnosis could be challenging in case of atypical radiographic presentation such as cortical destruction (6). Since misdiagnosis could result in inadequate overtreatment and cross-sectional imaging; thus histologic confirmation is necessary in case of atypical BPOP presentation (7).

We here reported a case of BPOP in a 34-year-old male that was presented with typical clinical, radiologic, and histologic findings. An antecedent trauma has been attributed to the etiology of BPOP (8). However, no history of trauma was recalled by the patient. The hand involvement is more frequent than feet, as was seen in our case (9). In one of the largest series of BPOP reports, the average age of the patients was 33.9 years, which was very close to the age of our patient (10). Pain and swelling are clinical features of BPOP that were present in our case as well (11).

The recurrence rate of as much as 50% has been reported following the marginal resection of BPOP within 2 years of resection (1). One-year-follow-up of our case revealed no sign of recurrence.

The most important differential diagnosis of BPOP is osteochondroma. The absence of continuity between the lesion and medullary cavity of the involved bone was suggested as the main radiographic finding for the differentiation of BPOP from osteochondroma (12), as indicated in this case. Even though, Rybak et al. recently reported the presence of corticomedullary continuity with the underlying bone, even in histologically proven BPOP cases (13). Thus this radiological finding could no longer be regarded as a reliable factor for distinguishing BPOP from osteochondroma. Cortical flaring at its junction with the lesion is commonly seen in osteochondromas. Such flaring was not present in the radiography of our case.

The cortical invasion, soft-tissue infiltration, and periosteal reaction are considered to be radiologic features present in parosteal osteosarcoma and absent in BPOP (12). However, cortical invasion and soft-tissue infiltration have been reported in BPOP less frequently (6). Therefore, BPOP must be histologically differentiated from parosteal osteosarcoma.

Peripheral chondrosarcoma is another main differential diagnosis of BPOP. A mass with ring-like or popcorn calcification in radiography is in favor of chondrosarcoma. Histologic differentiation of chondrosarcoma from BPOP is also performed by the presence of well-differentiated hyaline cartilage with lobular architecture, no mitosis in grade I chondrosarcoma, and increased cellularity and foci

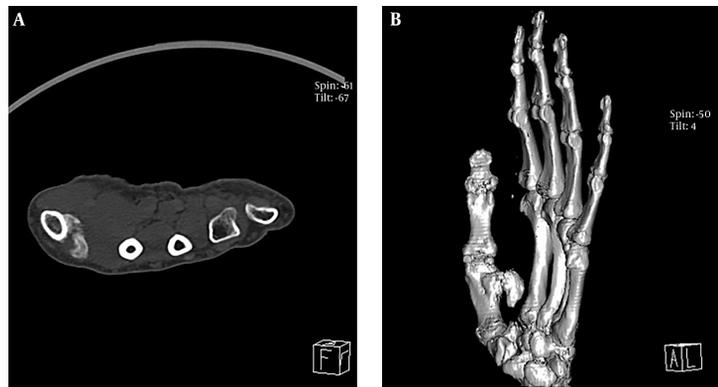


Figure 2. The 3D (A) and the axial (B) tomography of the left thumb demonstrating the BPOP lesion



Figure 3. Coronal T1-weighted MRI demonstrating a hypo-signal lesion without soft-tissue involvement

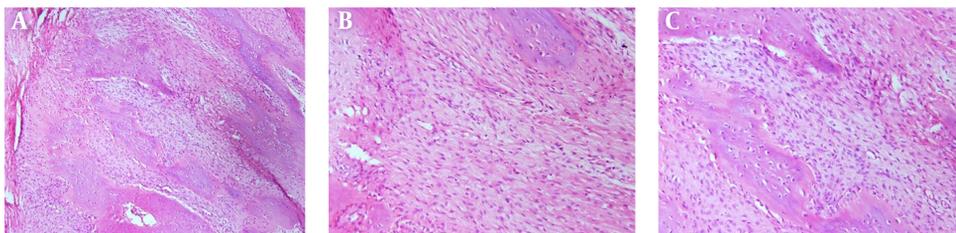


Figure 4. A, sheets of woven bone in a fibrous background (100x); B, hyper-cellular woven bone in the fibrous stroma (200x); C, The hyper-cellular proliferation of spindle cells with small pale nuclei and osteoid deposition (200x).

of necrosis in grade II variant (14).

Although BPOP usually presents with characteristics clinical and radiologic features, its atypical presentation is also reported. Thus its differential diagnosis from other

lesions, especially malignant tumors such as parosteal osteosarcoma and chondrosarcoma should be considered. To this aim, a combination of clinical, radiologic, and pathologic findings should be used to prevent misdiagnosis.

sis and overtreatment of the patients.

Footnotes

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