CASE REPORT

Bizarre parosteal osteochondromatous proliferation on a phalanx with periosteal erosion

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Summary  Bizarre parosteal osteochondromatous proliferation (BPOP) is an uncommon benign hand tumor with a high rate of local recurrence, marked proliferative activity, and an atypical histological appearance. The aim of this paper is to present a rare and illustrative example of BPOP with periosteal erosion. A 64-year-old male presented with a 10-year history of a mass, measuring approximately 3 cm in diameter, on the dorsal aspect of the right index finger. On physical examination, the mass was hard, indolent, and located at the level of the proximal phalanx. Roentgenograms displayed a soft tissue mass over the right index finger with erosion of the periosteum. Magnetic resonance imaging revealed a few well-defined lobulated tumors over the right index finger proximal interphalangeal joint with periosteal reaction. The soft tissue tumors were excised and found to have soft consistencies. Pathological findings demonstrated that the tumor was compatible with BPOP. BPOP is a benign but locally aggressive fibro-osseous mass yielding radiographic findings that bear striking clinical similarities to...
1. Introduction

Bizarre parosteal osteochondromatous proliferation (BPOP) is an uncommon benign tumor that usually presents as a bony swelling on the surface of a bone. It has a notably high rate of local recurrence (20–55%), marked proliferative activity, and an atypical histological appearance. BPOP is also called "Nora’s lesion," which was named after the pathologist who first discovered it at the Mayo Clinic in 1983. Nora et al reported 35 lesions found on a 50-year-old male patient, all involving the small bones of the hands and feet. In 1993, Meneses et al reported 65 cases of BPOP involving various sites, such as the hands, feet, long bones, and skull bones.

2. Case report

A 64-year-old man presented with a 10-year history of an irregular mass, measuring approximately 3 cm in diameter, on the dorsal aspect of the right index finger (Figure 1). He claimed to have no history of trauma. On examination, the mass was hard, indolent, and located at the level of the proximal phalanx. Movement of the interphalangeal joint was mildly restricted. Results of neurovascular examination were unremarkable, and all laboratory investigations were within normal limits. Roentgenograms revealed a soft tissue mass with faint calcification over the right index finger base, and the adjacent proximal phalanx displayed irregular erosion of the periosteum (Figure 2). Through magnetic resonance imaging, a few well-defined lobulated tumors were noted over the medial and lateral aspects of the proximal interphalangeal joint of the right index finger, along with bony erosion and periosteal reaction. The largest of these tumors was approximately $3 \times 2 \times 2 \text{ cm}^3$. The tumors appeared isointense to muscle on T1-weighted images (Figure 3A) and heterogeneously hyperintense on T2-weighted images. They exhibited heterogeneous contrast enhancement with gadolinium injection (Figure 3B).

Excision of the soft tissue tumors was performed using a dorsal longitudinal approach. The lobulated tumors were attached to the middle and proximal phalanges, and resected smoothly. Gross examination revealed a few lobulated tumors with soft consistencies and a maximum size of approximately $3 \times 2 \times 2 \text{ cm}^3$. The extensor tendon was intact, and an irregular cortical surface was noted near the proximal interphalangeal joint (Figure 4).

Pathological findings revealed irregular maturation of cartilage mixed with chondro-osteoid material, which had a bluish appearance in hematoxylin and eosin stain. Furthermore, numerous bizarre, binucleated chondrocytes were observed in the soft tissue. The tumor was compatible with BPOP (Figure 5).

3. Discussion

BPOP is a rare, reactive, mineralizing mesenchymal lesion that typically affects the surfaces of bones in the hands and feet, typically the proximal and middle phalanges as well as the metacarpal and metatarsal bones. The hands are affected four times more than the feet. The typical clinical presentation is a painless swelling that grows for months to years. Pain or skin rubor can be caused by the mass effect.

In typical radiological imaging, BPOP presents as a well-margined, ossified mass arising from the cortical surface. In general, the cortex is completely intact.

Figure 1 Preoperative pictures display an irregular mass measuring approximately 3 cm in diameter on the dorsal aspect of the right index finger.
Periosteal reaction and medullary involvement are typically absent in BPOP, which is associated with normal underlying bone and adjacent soft tissues.\(^4\,^6\,^8\,^9\) However, Barrera-Ochoa et al\(^1^0\) reported two atypical cases in 2012, each of which displayed the unusual radiographic feature of cortical destruction, which we also observed in the present case\(^1^1\). Additionally, in 2007, Rybak et al\(^5\) reported four cases of BPOP with radiographic evidence of medullary involvement. Thus, the presence of equivocal findings might cause diagnostic dilemmas regarding BPOP during radiological investigations. Radiology is not a reliable method for identifying or excluding BPOP.

Pathologically, BPOP has an atypical histological appearance, explaining the inclusion of the word “bizarre” in its name. Typical microscopic findings of BPOP include highly cellular, disorganized, and irregular cartilage, which is associated with the proliferation of bizarre-appearing fibroblasts, disorganized bone and spindle-shaped fibroblasts in the intertrabecular space, and bizarre binucleated chondrocytes of soft tissue.\(^4\,^1^2\) The bluish staining of the calcified cartilage matrix is a distinctive feature that has been interpreted as “blue bone.”

Clinically, BPOP presents with bony exostosis and demonstrates a fairly high recurrence rate of 29–55% within a 2-year interval.\(^1\,^1^3\) In the initial report by Nora et al,\(^1\) 18 of 35 cases (51%) recurred. Meneses et al\(^2\) and Dhondt et al\(^1^3\) reported recurrence rates of 55% and 29%, respectively. Surgical removal should be planned with close follow-up because of this high rate of recurrence after local resection. Although the benign lesion in the present case is prone to recurrence, aggressive management with a mutilating surgery might be avoided for this benign lesion.

In conclusion, BPOP is typically a well-defined mass that arises from the periosteal aspect of an intact cortex without medullary changes.\(^1\,^1^3\) However, atypical cases have also been reported, including the unusual radiographic feature of cortical destruction, which was observed in the present case. As the presence of equivocal radiological findings might cause diagnostic dilemmas regarding BPOP, radiological investigation is not a reliable method for identifying or excluding BPOP. Relying on pathological

Figure 2  X-ray images reveal a soft tissue mass with faint calcification over the index finger base (white arrows), and the adjacent proximal phalanx displays irregular erosion of the periosteum.

Figure 3  (A) Lesions appear isointense to muscle on the T1-weighted image. (B) Lesions appear heterogeneously hyperintense to muscle in the T2-weighted image. A few well-defined lobulated tumors are present (maximum size: \(3 \times 2 \times 2 \text{ cm}^3\); white arrow) over the medial and lateral aspect of the proximal interphalangeal joint of the right index finger. Bony erosion and periosteal reaction are also evident (white circle).
findings is still necessary for the correct differential diagnosis of this disease.

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References