Bizarre parosteal osteochondromatous proliferation of the phalanx

MG Agarwal, SG Prabhudesai, NA Jambhekar, A Duggal and A Puri

Department of Surgical Oncology, Bone & Soft Tissue Service, Tata Memorial Hospital,
Mumbai 400012. India

Correspondence
Dr Manish Agarwal
Associate Professor & Orthopaedic Oncologist
Bone & Soft Tissue Service
Tata Memorial Hospital
Mumbai 400012.
INDIA.
Fax  +  91 2 2414  6937
E-mail mgagarwal@yahoo.com

Abstract
Bizarre Parosteal Osteochondromatous Proliferation, are rare lesions presenting as bony
swellings on the surface of bone. Though simulating osteochondromas, or parosteal
osteosarcomas, they have a distinct radiological and histological appearance. The lesions
typically occur on hands and feet. The lesions are benign with a tendency to recur. We report a
lesion on the proximal phalanx of the little finger in a 21-year old student.
Introduction

Benign Parosteal Osteochondromatous Proliferation (BPOP) is a rare lesion involving the small bones of the hands, feet and less often the long bones. Nora et al first described them in 1983 [2]. Although, grossly these lesions resemble osteochondromas; histologically they exhibit marked proliferative activity and bizarre, enlarged and binucleate chondrocytes mimicking chondrosarcoma. These lesions were confused and included with juxta-cortical osteosarcoma in the early reports. The importance of these lesions is that their aggressive histological picture and high rate of local recurrence can lead to a mistaken diagnosis of malignancy.

Case Report

A 21-year old engineering student presented with a recurrent swelling of the left little finger (fig 1A) since four months. A similar swelling at the same site had been removed eighteen months back, in another institute. The patient gave no history of pain and there was no history of trauma preceding the swelling. On physical examination, a firm, discrete, immobile, non tender, bilobulated swelling could be felt on the antero-lateral aspect of the proximal phalanx of the left little finger. It was free from the overlying skin, which showed scar of previous surgery. There was no limitation of movement of the little finger.
Fig 1

A: Clinical photo at presentation. Note the swelling and scar of previous surgery.

B: Intraoperative photograph showing the lesion on the phalanx.

All laboratory investigations were normal. Plain radiography revealed two bony projections in the proximal phalanx of the left little finger (fig 2A). The distal bony projection appeared attached to the cortex while the proximal lesion appeared separate from it. No periosteal or cortical thickening was seen. CT scan showed the lesion to be surface lesion without medullary involvement or cortical erosion.
Fig 2

A: Preoperative X-ray showing ossified nodule on the surface of the phalanx (arrow) and a separate ossified mass in the soft tissue.

B: Postoperative X-ray showing the excised cortex.

C: Excised specimen as seen from the surface. The bony cortex is on the far and not visible in this picture.

The histology slides from his first surgery were reviewed at our institute (fig 3). They showed a cartilage cap with irregular maturation into bone. The cartilage was cellular with atypical chondrocytes. The accompanying ossification was seen as irregular trabeculae with a blue tinge especially at the interface with the cartilage. Spindle cells were seen between the trabeculae. The histological diagnosis offered was Bizarre Parosteal Osteochondromatous Proliferation.

At surgery the distal swelling appeared to be an osseous growth of two by three centimeters attached to the underlying proximal phalanx, but not involving the interphalangeal joint (fig 1B). It was excised along with the other nodule in continuity and with a part of the cortex of the
proximal phalanx (fig 2C). No connection with the medulla of the affected bone was apparent. The bony defect was not reconstructed (fig 2B). Postoperative stay was uneventful and the patient regained full range of movement. At a follow up of one year he was disease free and had full functional use of his hand.

**Fig3**:

*Photomicrograph showing the histology of the tumour. Note the bizarre appearance of the cartilage. Stain: H & E. Magnification: 100X*

**Discussion**

BPOP of bone (first described by Nora et al in 1983 [2] and called Nora’s lesion) are rare lesions. The commonest presentation is that of a bony swelling arising from the small bones of the hands and feet. Long bones such as ulna, radius, tibia, femur, fibula or humerus can be affected rarely [1]. Males and females are equally affected. It can occur in a wide age range but
is commonest in the third and fourth decades [1]. A history of trauma may be present in some cases. Pain is generally not present and symptoms are due to the swelling.

Radiologically these lesions appear to arise from the cortex without affecting it. There is no soft tissue mass beyond the mineralized portion. Grossly there is a cartilage cover with an underlying bony core. Histologically they consist of atypical cartilage with marked proliferative activity resembling Grade I or II chondrosarcoma [2], disorganized ossification in the cartilage, plenty of irregularly calcified osteoid with benign osteocytes, and actively proliferating but benign fibrous tissue.

The common differential diagnosis is osteochondroma, parosteal osteosarcoma, myositis ossificans or benign florid periostitis. Unlike in BPOP, osteochondromas are continuous with the medullary canal, and there is a flaring of the cortex at the point of attachment. The histological atypia of cartilage is far more with BPOP. The distal phalanx is never involved unlike subungual exostoses. Florid reactive periostitis described by Spjut and Dorfman in 1981 [3] usually presents with a mature or lamellated periosteal reaction and histologically has more mature bone and cartilage. Parosteal osteogenic sarcoma has been described in hands [4], but is more common in long bone. There are histologic similarities between this tumor and BPOP. In long bones the difference can be made by the lobular architecture of BPOP and the distinct blue tinctorial characteristic of the osteoid. Also BPOP is small, has irregular bony trabeculae and lacks invasion into adjacent muscle. In myositis the cartilage does not form a cap.

Zonation is seen with maturation and ossification first seen in the periphery. A periosteal reaction may be present. Over a period of time the lesion matures and diminishes.

It is still not clear whether BPOP is a neoplastic or reactive process. The frequent recurrences may support a neoplastic origin, however the histologic features showing a maturation of cartilage to bone suggest a reactive process [1].

The treatment is a simple excision. The recurrence rate is very high. Nora et al reported a 51%
rate of initial recurrence and a 22% of second recurrence. The majority of the recurrences occurred within two years of excision. A wide excision is probably curative.
References


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