

Case Report

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Bizarre parosteal osteochondromatous proliferation of humerus with unusual presentation: a report of one atypical case

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ABSTRACT

Bizarre Parosteal Osteochondromatous Proliferation (BPOP) as defined by Nora and colleagues in 1983 (called Nora lesion) is a rare benign, bony lesion and found mostly in metacarpals and metatarsals. BPOP in long bones are very rare. It is an exophytic outgrowth from the cortical surface comprising of bone, cartilage and fibrous tissue. The key radiological features to describe such lesions as calcified and osseous masses. We describe a case of BPOP in a 26 year old female involving the shaft of humerus. The radiological picture is a trabeculated osteolytic lesion. FNA revealed chondroma and histologic picture showed disordered pattern of cartilage, bone and fibrous tissue. This case is reported as the lesion is at a rare site (shaft of Humerus) unusual X-ray picture for the lesion and diagnostic histopathologic picture.

Keywords: BPOP, Osteochondroma, Trabeculated osteolytic lesion

INTRODUCTION

Bizarre Parosteal Osteochondromatous Proliferation (BPOP) as defined by Nora and colleagues in 1983¹ (called Nora lesion) is a rare benign, bony lesion and found in hand and feet. BPOP in long bones are very rare. About 160 cases of BPOP have been reported.² It is an exophytic outgrowth from the cortical surface comprising of bone, cartilage and fibrous tissue. The key radiological features to describe such lesions were 1) lack of corticomedullary continuity and 2) origin from periosteal aspect of an intact cortex.³ Grossly and X-ray picture resemble osteochondromas but histologically exhibit marked proliferation of bizarre, binucleate chondrocytes mimicking chondrosarcoma along with bone and fibrous tissue. This benign lesion of the bone might be misdiagnosed for malignant ones because of radiological pictures, atypical histopathologic appearance and high recurrence rates about.

CASE REPORT

A female patient of 26 year old admitted to the dept. of orthopaedics of SCB medical college, Cuttack, presented with slowly progressive solitary swelling of size 3x3 cm, on the shaft of humerus ovoid in shape, skin over the swelling is normal and pinchable with no sinus or scar over it. There was no history of trauma or pain preceding the swelling. On examination it was a firm, immobile, non-tender swelling at the lateral aspect of shaft of humerus.

X-ray revealed a lesion projecting from the cortical surface of humerus into the soft tissue. It was predominantly lucent, trabeculated, with a well-defined sclerotic rim merging with the underlying cortex. Endosteal surface is scalloped with no intramedullary component (Figure 1).



Figure 1: Trabeculated osteolytic bony lesion.

A provisional diagnosis of periosteal chondroma was made. FNA revealed mucoid material and cytosmear showed features of chondroma and suggested for biopsy and histopathologic evaluation. Tumour was excised; curettage and chemical cauterisation was done.

Gross

Gross received was multiple bits of greyish white tissue measuring 2x2x1 cm, gelatinous and gritty on cutting.

Microscopic

Microscopic examination of the tissue sections revealed cartilage, bone and spindle cells in a disordered pattern.

Cartilage appears hypercellular with enlarged and bizarre chondrocytes on a bluish background (Figure 2). Dense benign looking spindle cells are the proliferating fibrous tissue (Figure 3). Immature bone trabeculae stained mostly deep blue with haematoxylin and eosin (blue bone).

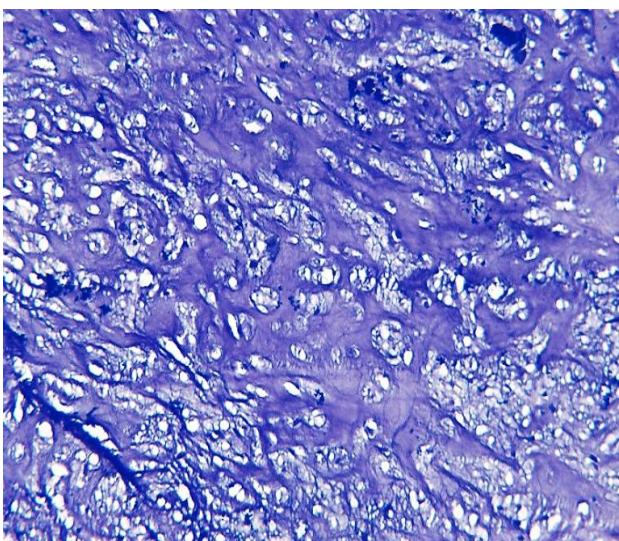


Figure 2: Micro section shows bizarre cartilage tissue 40x.

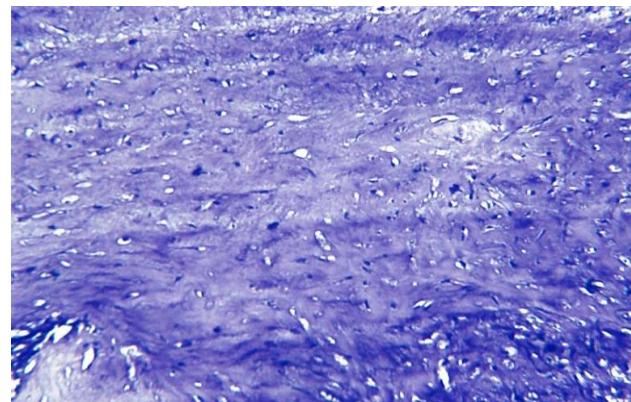


Figure 3: Micro section shows proliferation of fibroblastic cells.

Diagnosis

A diagnosis of bizarre parosteal osteochondromatous proliferation of humerus was made, a benign lesion.

DISCUSSION

BPOP of bone is an uncommon reactive mesenchymal bony lesion, called Noras lesion first described by Nora et al. in 1983. It typically affects the surface of small bones of hand and feet. Long bones like tibia, fibula, femur, radius, ulna or humerus affected rarely. It can occur in a wide age range, however commonest in the third and fourth decade. Male Female ratio is equal. The symptoms are mostly due to swelling. Recurrence rate is about 50%. Despite high recurrence and atypical histopathology appearance, no malignant transformation, metastases, or associated systemic diseases or death have been described so far in patients with BPOP.⁴ Here the long bone humerus is affected. The X-ray is typical with calcification and lytic but no cortical erosion or no corticomedullary continuity. FNA and H/P report was totally different to radiological picture. The histologic picture of extensive areas of benign bizarre cartilage; benign spindle cell proliferation along with blue bone formation gives a diagnosis of BPOP, a rare lesion. Excision was done, post up was uneventful. The recurrence was very common, usually within two years. A wide excision may be curative.

CONCLUSION

BPOP is a rare lesion of smaller bones however it can affect long bones also.⁵ The X-ray picture in almost all cases is a dense calcified and osseous masses but here in this rare case it is osteolytic. Clinical assessment and radiological diagnosis are not sufficient enough to treat a case of BPOP. Histopathologic evaluation should be performed for definite diagnosis and work up of all cases. Despite high recurrence and atypical h/p appearance, no malignant transformation, metastases, or associated systemic diseases or death have been described so far in patients with BPOP.

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Ethical approval: Not required

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