CASE REPORT

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Bizarre Parosteal Osteochondromatous Proliferation of the Humerus with Radiological Findings: A Case Report

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Abstract

Bizarre parosteal osteochondromatous proliferation (BPOP) is a rare, benign bone disease that primarily affects the metacarpals and metatarsals. We describe a 17-year-old male teenager with proximal humeral BPOP. It is a trabeculated osteolytic lesion on radiological examination. An excisional biopsy confirmed the diagnosis. Very few cases of BPOP in the long bones have been documented. It is an extension that emerges from the bone's cortical surface and is exophytic. Because the lesion was discovered in a rare location-the proximal diaphysis of the case is being reported. The gold standard for diagnosis is still the combination of radiographic and histological findings.

Keywords: BPOP, Bizarre parosteal osteochondromatous proliferation, Nora lesion, osteochondroma, osteolytic bone lesion

Introduction

Bizarre parosteal osteochondromatous proliferation (BPOP) is a relatively rare benign extraperiosteal osteochondroma-like proliferative lesion. Thirty-five cases involving the hands and feet were recorded when Nora et al. initially defined it in 1983 (1). Meneses et al. (2) identified 65 additional cases, with long bones being damaged in 17 of them.

Small bones in the hands and feet are most afflicted by this disorder, but long bones, vertebrae, skull, and jaw are also sporadically impacted (3). The hands account for most BPOP cases (55%), with feet coming in second (15%) and long bones in third (25%) (2). The second and third decades of life are when the incidence in adults peaks. It affects both men and women equally (3).

The normal presentation of BPOP is a firm, painful swelling that increases over time without causing harm. Diagnostic ambiguity arises from the rapid growth of this lesion and its similarity to malignant tumors like osteosarcoma and chondrosarcoma on imaging and histopathologic testing (2,3). The evaluation of both radiological and histological features is the basis for the diagnosis of BPOP (4). The cause of BPOP is currently unknown. The scarcity of BPOP means that the proof is scarce.

This study aimed to report this uncommon clinical condition and add to the body of knowledge regarding its management and aftercare.

Case Report

A 17-year-old boy presented with a 4-month history of swelling in his left shoulder. Upon investigation, a hard, painless swelling that was immobile was observed. There was no discomfort or limitation in motion. Trauma was not in the past.

The patient was first assessed using radiography. When an anteroposterior radiograph (Figure 1) revealed a well-defined radio-opaque bone lesion with exophytic extension in the proximal humerus, computed tomography (CT) was performed.

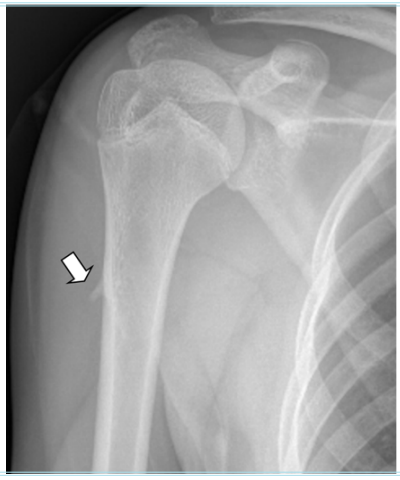

Figure 1. A well-defined radio-opaque bone lesion with exophytic extension in the proximal humerus (arrow)

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A smooth-circumscribed osseous lesion without medullary continuity was observed in the proximal diaphysis of the humerus on a non-contrast CT scan of the upper arm. The lesion extended exophytically from the cortex to the surrounding soft tissue (Figures 2A, 2B). In the soft tissue next to the identified lesion, no additional pathology was found (Figure 2C).

A pre-contrast T1-weighted magnetic resonance imaging (MRI) series revealed a bone lesion with bone-like density and a heterogeneous signal shift in the surrounding soft tissue. Soft tissue contrast uptake was used for the post-contrast MRI series (Figures 3A, 3B).

The main differential diagnoses were BPOP and parosteal osteosarcoma. An excisional biopsy of the lesion was performed to make a histological diagnosis. Histologically, the tumor surface contained fibrocartilaginous tissue with considerable cellularity. The cells varied in size, with some being binucleated. The basal area consisted of juvenile bone trabeculae with significant osteoblastic activity. The cells demonstrated abnormal mitosis but no cytologic atypia. Thus, the diagnosis of BPOP was confirmed.

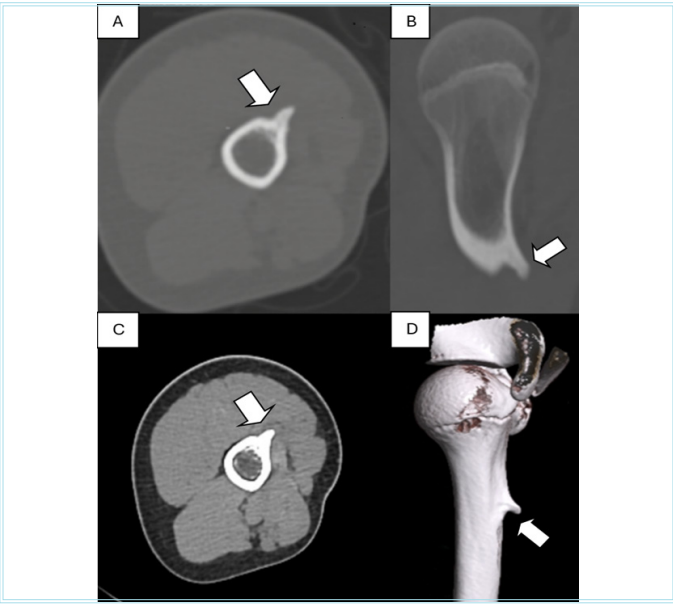

Figure 2. On a non-contrast computed tomography (CT) scan of the upper arm. An axial and coronal image displays a well-defined exophytic bone lesion in the bone window that lacks medullary continuity (A, B, arrow). The axial image shows no other pathology in the soft tissue adjacent to the lesion (C, arrow). We observe an exophytic bone lesion emanating from the diaphysis in the 3-dimensional-CT humerus image (D, arrow)

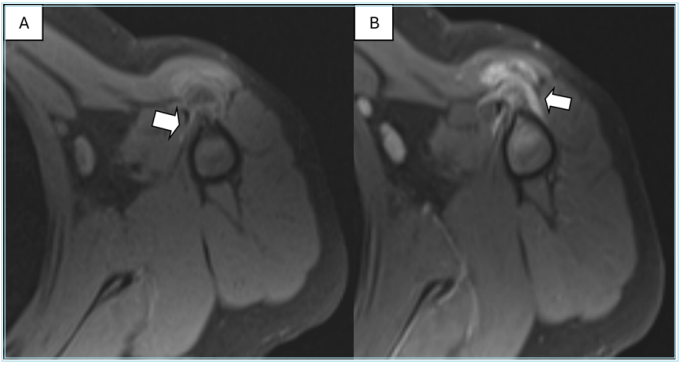

Figure 3. An exophytic bone lesion and a heterogeneous signal shift in the surrounding soft tissue are visible on pre-contrast T1-weighted magnetic resonance images (A, arrow). The lesion enhances similarly with the adjacent bone. Increased soft tissue contrast enhancement is seen in post-contrast magnetic resonance imaging series (B, arrow)

Discussion

BPOP of bone is an uncommon reactive bone lesion known as Noras lesion, which was initially identified by Nora et al. (1) in 1983. It primarily affects the bones of the hands and feet. Long bones, including the tibia, fibula, femur, radius, and ulna, are rarely impacted (2). It can affect people of all ages, but it is most common in the second and third decades. The ratio of men to women is equal. The most common cause of symptoms is edema (3). In our case, the swelling was painless, as described in the literature.

As far as we know, in the English literature, there have been around 200 cases of BPOP recorded to date. In addition to its rarity, BPOP is less common in long bones (4).

Even more uncommonly, one of the long bones, the humerus, was afflicted. Excision was performed, and the follow-up proceeded well.

According to the most extensive radiology-based study to date, BPOP is a well-defined mass of heterotopic mineralization arising from the periosteum, with an intact cortex and no medullary alterations (5). Periosteal new bone growth is not observed in BPOP. The lack of continuity between the lesion and the bone's medullary cavity is an essential radiographic finding that distinguishes BPOP from osteochondromas. In addition, there is no cortical hypertrophy (6).

Although BPOP has distinct clinical and histological features, it may be mistaken for other benign and malignant diseases. Because of its parosteal location, BPOP must be distinguished from parosteal osteosarcoma, which is uncommon in the hands and feet (7). The lack of cellular atypia distinguishes this disease from osteosarcoma. The lesion's surface position and cartilaginous component may lead to confusion with osteochondroma. Osteochondromas are relatively uncommon in the tiny bones of the distal extremities (8). They have typical continuity with the medullary canal, and the cartilage displays no symptoms of atypia.

Rybak et al. (9) described four cases of BPOP with corticomedullary continuity with the underlying bone on imaging, which was verified by pathological diagnosis. Thus, radiologic characteristics alone cannot define BPOP, according to Rybak et al.'s (9). For a conclusive diagnosis, a histopathological investigation should be performed.

In histological sampling, another important marker for the diagnosis of BPOP, there are three components of BPOP: cartilage, bone, and spindle cells. Cartilage usually forms a cap; less frequently, it is arranged in lobules separated by dense fibrous tissue with irregular maturation into bone (endchondral ossification) and spindle cells in the background. Cartilage is hypercellular and contains large chondrocyte. Binucleated cells are common, and hyperchromasia and cytologic atypia are absent. Mitotic figures are common but do not show atypia (2).

The bone lesion's radiologic and histopathologic characteristics in our instance matched those reported in the literature.

The rate of recurrence is approximately 50%. Thus far, no malignant transformation, metastasis, associated systemic disease, or death has been reported in patients with BPOP, despite the high recurrence and

emergence of aberrant histology. Given the frequency of recurrence, a broad excision would be beneficial (10,11).

Conclusion

In conclusion, long bones can also be affected by BPOP, which is a rare lesion of small bones. The gold standard for diagnosis is still the combination of radiographic and histological findings.

Ethics

Informed Consent: Written consent was obtained from the patients and their relatives who participated in this study.

Authorship Contributions

Surgical and Medical Practices: E.B., Concept: K.B.M., Design: E.B., Data Collection or Processing: K.B.M., Analysis or Interpretation: K.B.M., Literature Search: E.B., Writing: E.B., K.B.M.

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