Bizarre parosteal osteochondromatous proliferation of the ring finger: a case report and review of the literature

Dawood Jafari, MD,1 Hamid Taheri, MD,2 Hooman Shariatzadeh, MD,3 Khodamorad Jamshidi, MD4, Alireza Pahlevansabagh, MD5

Department of Hand Surgery, Shafa Yahyaian Hospital, Iran University of Medical Sciences, Tehran, Iran.

Abstract

Bizarre parosteal osteochondromatous proliferation (BPOP), also known as Nora’s lesion is a rare osteocartilaginous lesion composed of a disorganized mixture of cartilage, bone, and fibrous tissue. In this article we report a case of BPOP arising on the proximal and middle phalanx of ring finger in a 31 year-old woman. The clinical, radiographic, MR imaging and histopathologic findings of it are described. The symptoms regressed spontaneously in 2 month after incisional biopsy.

Keywords: benign tumor, phalanges, bizarre parosteal osteochondromatous proliferation, Nora’s lesion.

Introduction

In 1983, Nora et al. [1] described a rare tumor in the hand and foot and called it bizarre parosteal osteochondromatous proliferation (BPOP). BPOP is a rare osteocartilaginous lesion of bone composed of a disorganized mixture of cartilage, bone, and fibrous tissue [2]. It arises most commonly in the small bones of the hands and feet but also has been reported in other bones, including the long bones of the upper and lower limb [1, 3]. BPOP is thought to be a reactive lesion, closely related to florid reactive periostitis, subungual exostosis, and turret exostosis [4]. In the literature this disorder has been referred to by a confusing variety of names; pseudomalignant osseous tumor of the digits [5], extraskeletal localized non-neoplastic bone, cartilage formation or myositis ossificans. Careful histologic examination is usually necessary to distinguish this benign lesion from other disorders, due to the fact that this disorder has specific pathological characteristics. Clinical course is typically benign and local resection is the definitive treatment. However, recurrences have been reported that required more aggressive management. This report is unique because of spontaneous regression of BPOP affecting the ring finger after incisional biopsy of the lesion over the time.

Case report

A healthy 31-years old right-handed housewife presented with a 3-months history of a progressive growing painful mass on the proximal and middle phalanx of her left ring finger. Her past history was notable for superficial second degree burning injury of this finger 6 months ago. The pain worsened gradually during this 3 month. Physical examination revealed a mass adjacent to the proximal and middle phalanx

1&2&4. Associate Professor of Orthopaedic Surgery, Iran University of Medical Sciences, Tehran, Iran
3. Assistant Professor of Orthopaedic Surgery, Iran University of Medical Sciences.
5. Corresponding author, Hand Surgery Fellow, Shafa Yahyaian Hospital, Baharestan Sq, Tehran, Iran. Tel: +9821 33542022.
E Mail: pahlevansabagh@yahoo.com
with subcutaneous swelling (Fig. 1). The tumor was remarkably hard and seemed to be fixed to the skeleton of the finger. The overlying skin was pink and the tumor was severely tender. The PIP joint range of motion was limited compared to the right ring, whilst motion of the metacarpophalangeal joint was unaffected. The sensibility and vascularity was normal. During the patient's first visit to our hospital, diffuse swelling, heat, erythema and tenderness around the ring all indicated a local inflammation. Signs of infection, such as fever, cellulitis or lymphadenopathy, were absent. Laboratory study results, including C-reactive protein, complete blood count, erythrocyte sedimentation rate and serum levels of calcium, phosphorus, alkaline phosphatase and uric acid were within normal range. The radiographs (Fig. 2) revealed increased soft tissue density surrounding the proximal and middle phalanx; the cortex was intact and fine periosteal reaction was obvious. Magnetic resonance imaging (MRI) indicated a mass surrounding the proximal and middle phalanx. This mass was iso-intense on T1-weighted images, high-intense on T2-weighted images. Soft tissue around the lesion also showed high intensity on T2 images, indicating edema. The clinical and radiological symptoms were interpreted as "malignant tumor." The finger was explored and an incisional biopsy was performed. The tumor was adhering to the bone and extensor tendon (Fig. 3).
Macroscopically the mass appeared to be soft and creamy in color. Specimen for light microscopy were fixed in 6% formalin and embedded in paraffin. The 5 mm sections were stained with hematoxylin and eosin. The lesion consisted of a mixture of cartilage, bone and fibrous tissue. The cartilaginous fragments were of moderate cellularity. The chondrocytes showed mild nuclear atypia. No mitotic figures were found. The immature bone trabeculae were unevenly calcified and lined by prominent but cytologically benign osteoblasts. The intermixed fibrous tissue showed no abnormalities (Fig. 4). The histology was described as a BPOP. Aerobic and anaerobic cultures were negative. After incisional biopsy and under careful observation of the clinical and radiological features, the pain disappeared in 2 month without any administration of medicine and the mass spontaneously decreased in size as seen on photography (Fig. 5). Three months after the first MRI, another MRI study was performed for re-evaluation. The mass surrounding the phalangeal bone was markedly reduced in size, both T1- and T2-weighted images showed low intensity indicating hardening of the lesion, and few inflammatory signs were seen in the soft tissue around the bone. Twenty months after onset, no more clinical and radiological symptoms were observed and the patient had regained full range of interphalangeal motion. To evaluate any recurrence, clinical examinations and X-rays will be performed every 3 months.

**Discussion**

In 1981 Spjut and Dorfman [6] reported cas-
es of reactive florid periostitis in hand and feet
phalanxes. All their cases showed abundant os-
teoid production from a proliferative fibrous
stroma. Because periostea was involved in all
cases, these investigators coined the term
"florid reactive periostitis (FRP)" to more accu-
rately describe the pathogenesis of the lesion.
Florid reactive periostitis is more frequent in
the second and third decades of life [7]. There
are more women than men suffering from florid
reactive periostitis [8]. These tumors are usually
found in hand phalanxes. Brien et al [9] de-
scribe a case of a florid reactive periostitis of the
tibia as an atypical case of tubular bones.

In 1983 Nora et al [1] described cases of
bizarre parosteal osteochondromatous prolifer-
atations. Although the lesions they described
showed some histological similarities to those
reported by Spjut and Dorfman [6], there was a
pronounced tendency to local recurrence, ap-
proximately 50% of cases, and they considered
this to be a distinct process from the one
described by Spjut and Dorfman [6]. Male and fe-
males were equally affected and the age range
was very broad [10]. BPOP usually arises in the
small tubular bones of the hands and feet but
has very rarely been reported in other bones, in-
cluding the humerus, radius, ulna and clavicle
[2,3,11]. So far many synonyms such as myosi-
tis ossificans, parosteal fasciitis, pseudosarco-
matus fibromatosis, and fasciitis ossificans have
been used to describe FRP and BPOP [1,6,
12,13]. FRP is a typical tumor of soft tissue of the
hand with a membranous type of ossifica-
tion: direct ossification of fibrous tissue with-
out the cartilaginous stage. Enchondromal ossi-
fication can be seen in BPOP [10].

Yuen et al. observed considerable overlap be-
tween lesions identified as FRP and BPOP and
also suggested that FRP could progress to
BPOP. They therefore proposed the term "pro-
liferative periosteal process of phalanges" for
such non-neoplastic reactive changes accom-
panied by bone formation (D20).

Typical clinical findings are soft tissue tu-
mors, differing degrees of pain and swelling,
and a skin erythema of several month evolu-
tions.

When a soft tissue swelling occurring in a
small tubular bone of the hand as in our case is
encountered, infection, tumor, fracture or in-
flammation must be considered as possible lo-
cal causes and secondary pulmonary hyperos-
tosis as a systemic cause. Fracture is a well-
known and frequently encountered problem in
the hand and its presence or absence can be de-
termined by examining the patient's history.
Two other causes of soft tissue swelling, infec-
tion and inflammation, may be excluded by
physical examination, laboratory studies and
radiological findings. Tumor, parosteal and pe-
riosteal osteosarcoma, which are rare in small
tubular bones, also must be included in a differ-
ential diagnosis.

Histologically and radiologically there is an
overlap between BPOP, florid periostitis,
parosteal osteosarcoma and osteochondroma
[4]. In BPOP there is a disorganized prolifer-
ation of fibrous tissue, cartilage and bone. Con-
trary to osteosarcoma, no atypical fibrous spind-
le cells are present. The chondrocytes are
bizarre, enlarged and sometimes binucleated.
Ossification is unlike osteochondroma or a sub-
ungual exostosis, very irregular and has an ap-
pearance similar to callus tissue. In addition, di-
rect metaplasia from fibroblast to osteoblast
(membranous ossification) can be observed,
similar to florid periostitis. In the latter, howev-
er, cartilage is absent. In contrast to florid pe-
riostitis, periosteal reaction is absent in BPOP
[4,14]. The most common differential diagno-
sis is osteochondroma. BPOP is distinguished
histologically by the absence of organized car-
tilage and endochondral ossification, as well as
the presence of areas of fibroblast proliferation
and fibrocartilaginous and fibroosseous meta-
plasia [1,2]. Radiologically there is no contin-
uity with the cortex of the medullary canal in
BPOP. In osteochondroma the cortex flares out
and there is communication with the medullary
canal [10]. Our case did not present any evidence of cortical bone erosion. The authors think that resonance magnetic scans do not yield more data than clinical and radiographic evaluation, although they may allow us to define better tumor location and infiltration of neurovascular bundles. It may also be of help in assessing recurrence after surgical excision.

Due to painful swelling and bone formations with the radiological appearance of tumorous lesions, some surgical treatment including marginal resection and ray amputation is often selected for these lesions [7]. BPOP, however, is known to recur frequently, in over 50% of cases [15]; this feature, in addition to the finding of a number of cytogenetic abnormalities, including at (1;17)(q32;q21) translocation, has led to the suggestion that BPOP represents a neoplastic rather than a reactive lesion [16]. Malignant transformation, except in 1 case of fibrosarcoma arising in association with BPOP [17], has not been reported and is not considered typical of this lesion.

In our case, after incisional biopsy the pain disappeared in 1 month and the mass spontaneously decreased in size and after twenty month no recurrence was observed. Therefore in view of the possibility of this disease becoming spontaneously regressive, careful follow-up may be preferable to immediate surgery.

Due to high rate of recurrence, clinical examinations and X-rays were performed every 3 months. The association with scar of burning was not reported. We could not find any relationship between the two pathologies.

References