Skeletal Radiol (2001) 30:192–198

ARTICLE

Murali Sundaram
Lihuan "Lisa" Wang
Mitchell Rotman
Richard Howard
Alesia P. Saboeiro

Florid reactive periostitis and bizarre parosteal osteochondromatous proliferation: pre-biopsy imaging evolution, treatment and outcome

Received: 21 August 2000
Revision requested: 8 December 2000
Revision received: 26 December 2000
Accepted: 26 December 2000

M. Sundaram, M.D. (✉) · L. Wang, M.D.
Department of Radiology,
St. Louis University Health Sciences Center,
3635 Vista at Grand, St. Louis,
MO 63110-0250, USA

M. Rotman, M.D. · R. Howard, D.O.
Department of Orthopedic Surgery,
St. Louis University Health Sciences Center,
St. Louis, Missouri, USA

A.P. Saboeiro, M.D.
Division of Plastic Surgery,
St. Louis University Health Sciences Center,
St. Louis, Missouri, USA

Abstract *Objective.* To report on the imaging evolution of florid reactive periostitis (FRP) and bizarre parosteal osteochondromatous proliferation (BPOP) of the phalanges of the hands from prospective diagnosis to operation and on postsurgical outcome.
Design and patients. Three patients (2 female, 1 male; age range 11–34 years) presented with a swollen digit of the hand. Following presumptive radiographic diagnosis of FRP, they were closely observed both clinically and radiographically until operation. All three patients had radiographs of the involved digit, and one patient had an MR imaging examination. The interval between presumptive diagnosis and operation ranged from 2 to 8 months. Following operation, the patients have been clinically followed for 9–13 months (mean 10 months).
Results. In each of the patients, maturing of periosteal reaction without bone destruction was observed within 1–2 weeks of the presumptive diagnosis of FRP. Periosteal reaction was initially minimal in relation to the extent of soft tissue swelling and subsequently became more florid. In one patient, the lesion ossified, became adherent to the phalanx, and had an "osteochondromatous" appearance. In another patient, periosteal reaction was seen on both sides of the phalanx with an intact phalanx. In the sole patient who had MR imaging, edema was seen in the phalanx distal to the symptomatic site and the metacarpal proximal to the symptomatic site.
Conclusions. Close clinical and radiographic correlation permits an accurate pre-biopsy diagnosis of FRP. The first follow-up radiograph taken within 2 weeks usually provides reassurance of the accuracy of the diagnosis. FRP may progress to BPOP. Arbitrary antibiotic treatment can be avoided, and a planned surgical approach can be adopted.

Keywords Florid reactive periostitis · Bizarre parosteal osteochondromatous proliferation · Phalanges · Hand · Radiographs · MRI

Introduction

The term "florid reactive periostitis" (FRP) was coined almost 20 years ago to describe an aggressive periosteal reaction associated with soft tissue swelling of benign etiology and mimicking osteosarcoma [1]. This entity had been previously described as parosteal fasciitis [2] and parosteal (nodular fasciitis) [3]. Two years after the report on FRP, another reactive bone-producing lesion of the small bones of the hands and feet under the title of bizarre parosteal osteochondromatous proliferations of the hands and feet (BPOP) was described [4]. FRP and BPOP have been considered reactive lesions mimicking infection and tumor and usually have been considered as separate and distinct, although with increasing experience some overlapping histological features have been

noted [5]. A hypothesis unifying the nosological distinctions between FRP, BPOP and turret exostosis was proposed 8 years ago without imaging findings to prove it [6]. It would seem that this could only be done by allowing the lesion to take its course.

This paper describes three patients in whom the presumptive diagnosis of FRP was accepted by the referring orthopedic hand surgeons, who withheld biopsy and closely followed the patients until bone reaction matured, after which excision was performed (Figs. 1–3). In certainly one and possibly two patients the lesion clearly progressed from FRP to BPOP, illustrating that they represent a continuum of the same entity (Figs. 1, 2).

Case reports

Case 1

An 11-year-old afebrile, right-hand-dominant Caucasian female was admitted from an outside hospital with the chief complaint of redness, pain and swelling of the right fourth digit. The family reported that the patient had fallen while playing basketball 1 month earlier without injury. About 2 weeks later, the patient noted acute onset of pain and swelling of the proximal phalanx of the fourth digit. Radiographs at the outside hospital (April 2, 1999) showed periosteal reaction (Fig. 1A). The patient was splinted for treatment of a presumptive healing fracture.

However, on April 14, the patient's pain and swelling increased and she was admitted to the hospital for osteomyelitis treatment. The initial physical examination demonstrated a right fourth digit with proximal phalangeal erythema and swelling on the ulnar aspect. Digital flexion was limited secondary to pain and swelling. Complete blood count and sedimentation rate were normal. The orthopedic surgeons were considering open drainage versus soft tissue aspiration to determine the etiology of the swelling.

Upon review of the outside radiographs (Fig. 1A) with the more recent radiographs (April 19; Fig. 1B, C), the radiologist felt that the finding of the increasing mature periosteal reaction of the right fourth digit with normal cancellous bone most likely represented FRP. With this diagnosis, the patient was discharged home with a splint and no antibiotics. Short clinic follow-up with hand radiographs revealed increasing soft tissue swelling and periosteal reaction involving the medial and lateral portions of the fourth proximal phalanx with scalloping along its lateral inferior margins (May 1; Fig. 1D, E).

On May 14, even though the radiographic diagnosis remained FRP, the orthopedic hand service was concerned about a tumor because of the massive swelling (Fig. 1F). Subsequently, a small percutaneous closed biopsy was obtained and proved to be reactive periostitis. Radiographs from July 12 and December 2 showed a dramatic increase in ossification with a growing bony mass confluent with the medial aspect of the proximal phalanx of the right fourth digit, reminiscent of BPOP. Following the radiograph of July 12, it was decided to remove the mass electively during school recess for Christmas. The examination of July 12 showed no significant change from that of December 22 (Fig. 1G, H).

On December 22, excision of the digital mass was performed. The mass enclosed the ulnar digital artery and dorsal sensory branch of the ulnar nerve. Gross pathology showed a 4.0×2.5×1.5 cm oval, dome-shaped, tan-gray bone partially covered by tan-gray soft tissue. Histological examination revealed a benign bony lesion with trabeculae of lamellar and woven bone consistent with BPOP. The medullary space was fibrotic. Focal chondral differentiation was present. A structure very similar to a simple bone cyst was found within the bony lesion.

Follow-up radiographs on April 3, 2000, 3 months following removal of the bone mass from the proximal phalanx, revealed no evidence of soft tissue mineralization or recurrence (Fig. 1I). The patient denied any tenderness and had a significantly increased range of motion. Clinical follow-up 6 months after surgery showed normal function of the digit.

Case 2

A 34-year-old right-hand-dominant Caucasian female with a 13-year history of Behçet's disease complained of

Fig. 1A–I Case 1. An 11-year-old afebrile, right-hand-dominant female with a swollen right fourth digit was referred for confirmation of osteomyelitis and intravenous antibiotic treatment. **A** Fine periosteal reaction adjacent to the proximal medial cortex of the proximal phalanx led to a diagnosis of a healing fracture and the finger splinted (April 2, 1999). **B**, **C** Increase in swelling and periosteal reaction, more pronounced laterally, resulted in the patient being referred to our institution (19 April, 1999). Note there is no cortical destruction, although there is minimal scalloping medially and there is also a fine periosteal reaction laterally. At this point in the patient's evaluation, the diagnosis of florid reactive periostitis (FRP) was offered and early follow-up advised. **D**, **E** Examination 12 days later shows maturing and more profound periosteal reaction medially and laterally without cortical destruction. At this point in the patient's evaluation, the radiographic diagnosis was firm in that the patient had FRP (May 1, 1999). **F** Two weeks following **D** and **E**, the patient returned for routine clinic follow-up and marked soft tissue swelling of the finger caused clinical concern for an underlying soft tissue abnormality. Note again the lesion shows more profound bone medially conforming to bizarre parosteal osteochondromatous proliferation (BPOP) with periosteal reaction laterally but no bone destruction (May 14, 1999). **G**, **H** Large, bulky, ossified "osteochondromatous" lesion is adherent to the medial cortex which it has scalloped. Periosteal reaction laterally is solid. The radiographic appearance is that of BPOP (July 12, 1999). **I** Three months following resection of the mass, slight residual swelling is noted and function had returned to normal (April 3, 2000)

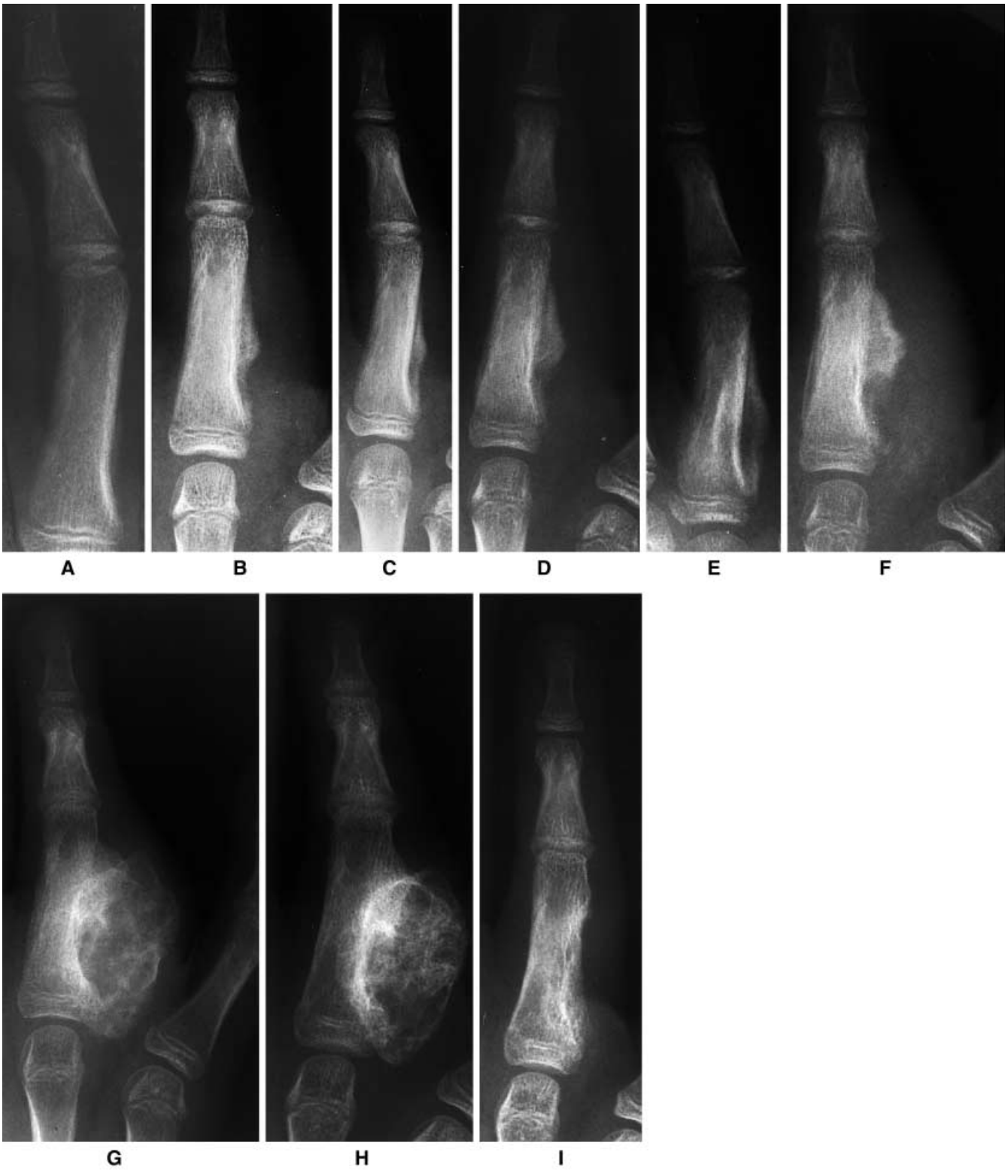

Fig. 1A–I Legend see page 193

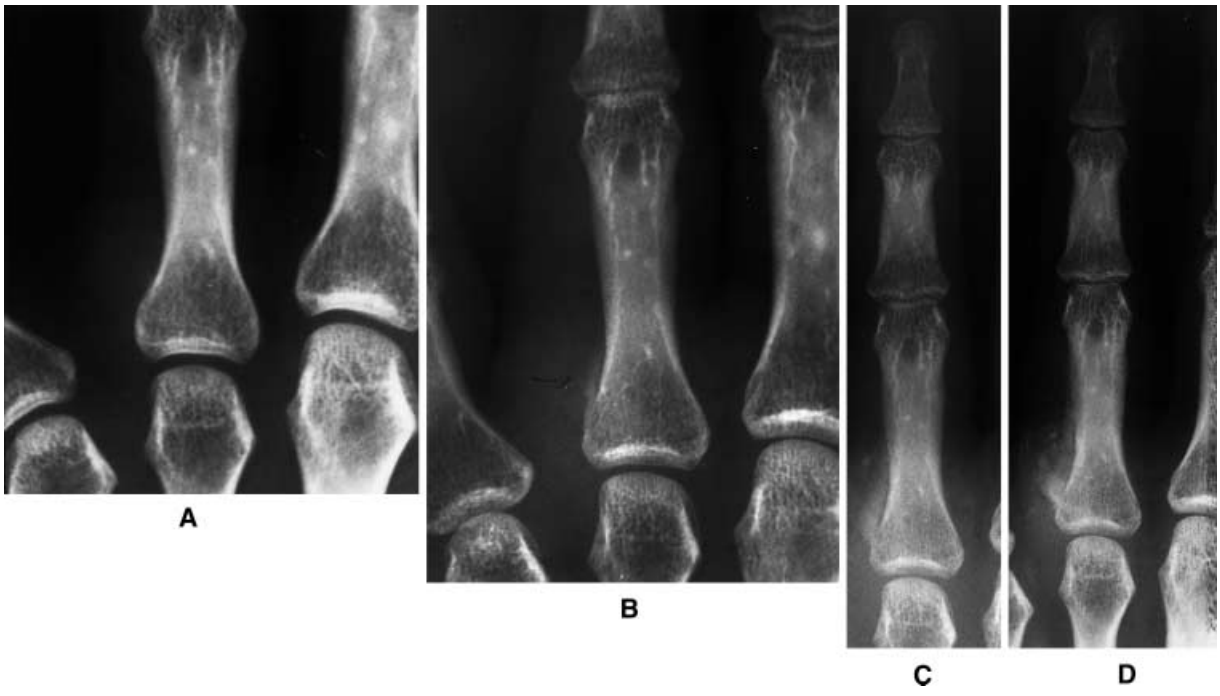

Fig. 2A–D Case 2. A 34-year-old female presented with soft tissue swelling at the base of the fourth digit of the left hand. **A** Soft tissue swelling of the medial aspect without underlying bony abnormality. The patient was treated with cortisone following this clinical and radiographic evaluation (July 2, 1999). **B** Subtle periosteal reaction that appears somewhat solid is noted at the base of the proximal phalanx medially. The patient was referred to our institution for evaluation of a bone tumor (July 6, 1999). **C**, **D** Radiographs of the digit taken 2 weeks and 4 weeks later (July 19 and August 2, 1999, respectively). Increasing, maturing bone production is noted medially without cortical destruction, consistent with an early BPOP lesion

a 1-month history of an erythematous and swollen left fourth digit with increasing difficulty in making a fist. Initially, the patient's primary care physician treated the symptoms as tendonitis with parenteral cortisone. When radiographs from July 2, 1999 revealed periosteal reaction of the left fourth digit, the patient was referred to the hand specialist for investigation of a possible hand tumor (Fig. 2A).

The physical examination revealed an erythematous and swollen left fourth digit with intact sensation and decreased range of motion. Radiographs taken at the first consultation visit on July 6, revealed soft tissue mineralization on the ulnar aspect of the left fourth proximal phalanx with subtle solid periosteal reaction (Fig. 2B). The radiographic impression was that of FRP with the recommendation of short-interval follow-up with repeat radiographs instead of a biopsy.

Sequential radiographs from July 19 and August 2 demonstrated increasing ossification and maturation of the periosteal reaction adjacent to the medial aspect of the left fourth proximal phalanx (Fig. 2C, D). On August 27, the mass was resected when the mass appeared localized.

During resection, the bony mass was found wrapped around the ulnar digital nerve. Gross pathological examination revealed four tan-white, firm, irregularly shaped tissue fragments measuring 3.5 cm×2.5 cm×0.7 cm in total. Histological examination showed fibrous connective tissue, osteoid with osseous metaplasia, and metaplastic cartilage consistent with reactive periostitis.

On the 1-month follow-up radiograph of September 30, no recurrence was identified. The patient had a full range of motion with significant decrease in tenderness. At 13-month follow-up, the patient had no problems referable to the digit.

Case 3

A previously healthy 19-year-old, right-hand dominant Caucasian male complained of a swollen and painful left third digit for 2 months. The patient denied a recent history of trauma. After being treated with antibiotics, the patient was referred to the orthopedic hand surgeon for biopsy when radiographs revealed a periosteal reaction.

The physical examination revealed a sausage-like left third digit with swelling extending to the metacarpophalangeal joint and involving the flexor sheath. Range of motion was 20° at each digit joint secondary to pain. The initial radiograph from September 29, 1999 demonstrated diffuse soft tissue swelling of the third digit with periosteal reaction on the medial and lateral sides of the proximal and medial aspect of the middle phalanx

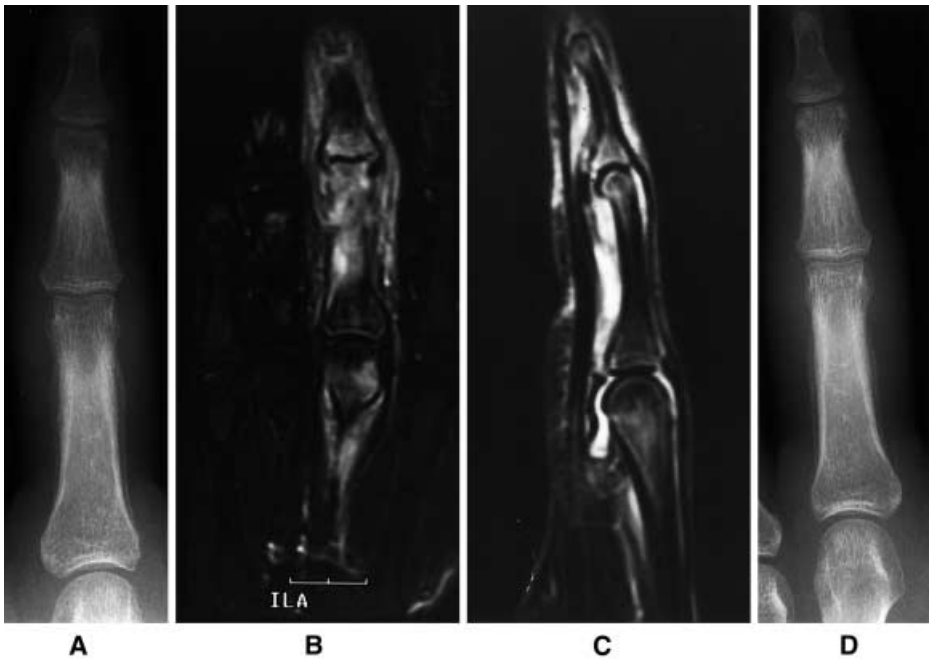

Fig. 3A–D Case 3. A 19-year-old right-hand-dominant male with a swollen, painful left long digit of 2 months' duration. **A** Periosteal reaction noted along the entire length of the shaft of the proximal phalanx medially and laterally, surrounding the middle phalanx. The radiographic diagnosis was felt to be consistent with florid reactive periostitis (September 29, 1999). **B**, **C** STIR coronal and sagittal MR images show considerable edema-like fluid between the tendons and bones with some edema in the subcutaneous soft tissues and in the metacarpal heads and phalanges. There is no joint effusion and no evidence of bone destruction (October 6, 1999). **D** Examination 4 weeks following initial presentation shows no change in the periosteal reactions with intact underlying phalanx and persisting soft tissue swelling (October 27, 1999)

(Fig. 3A). The cortex was intact. The radiographic findings were felt to be compatible with FRP.

With the radiological diagnosis of FRP, the patient was placed on Indocin and followed with short-interval hand radiographs and clinic visits. On October 6, MR imaging of the hand revealed T2 prolongation within the marrow of the third proximal phalanx, base of the middle phalanx, and the head of the third metacarpal bone was increased. Considerable edema-like fluid was noted between the tendons and bones with less edema in the soft tissues dorsal and volar to the tendons (Fig. 3B, C). A follow-up radiograph on October 27 showed no significant change (Fig. 3D). On December 7, the mature periosteal bone formation was excised.

Gross pathological examination showed one tan-white, irregularly shaped rubbery tissue fragment measuring 3.0×1.7×0.5 cm. Histological examination demonstrated fibro-adipose tissue with focal fibroblast proliferation. A small fragment consistent with synovium had mild chronic inflammation.

Follow-up radiographs after excision showed periarticular osteopenia with solid incorporated mature periostitis in the left third proximal phalanx. At most recent follow-up, this patient still had some swelling but no pain or radiographic evidence of mineral in soft tissues.

Discussion

We have not been able to find any series where the diagnosis of FRP or BPOP was presumptively made, biopsy withheld, and operation delayed. This is likely because of the performance of a biopsy or excision at the time of discovery, usually to exclude a malignant or infectious disease process. The rationale for delaying an operation in our patients was to determine whether the lesion would mature and ossify for ease of removal, as is often the management approach with heterotopic ossification in the lower extremities.

Each of our patients presented with a hugely swollen finger. In the two adults, the swelling had been present for 2 months with unsuccessful treatment elsewhere, and the child had had the swelling for 2 weeks. Referral to the hand surgeon in all three cases was prompted by the appearance of a periosteal reaction and for treatment of either osteomyelitis or "a tumor".

The clinical and radiographic features that influenced us in primarily suggesting the diagnosis of FRP were the presence of soft tissue swelling clearly out of proportion to the extent of periosteal reaction and an intact bony phalanx which spoke against either a bone tumor or osteomyelitis and suggested a reactive lesion such as FRP. A second feature that was perhaps the

most reassuring in making this diagnosis was the rapid progression of periosteal bone production with an intact cortex and no change in soft tissue swelling (Fig. 1B, C, Fig. 2C, D). In one patient, the presence of periosteal reaction developing on both sides of the phalangeal cortices with an intact cortex made FRP the most plausible diagnosis (Fig. 3A). From a clinical and imaging standpoint, it is during this stage of marked soft tissue swelling and aggressive periosteal reaction that the lesion resembles a malignant disease process either from bone or the so-called pseudo-malignant osseous soft tissue tumor [7]. In the single patient who had MR imaging, the only additional information provided was the presence of edema in the soft tissues and bones but not in the joint spaces proximal and distal to the site of maximal clinical and radiographic abnormality. We do not believe that MR imaging is required to make the diagnosis of FRP, although it could be utilized if the clinical and radiological appearances raise the possibility of a septic arthritis.

Case 1 is of interest in that over a 6-month period her lesion progressed from FRP reactive periostitis to a massive bony tumor-like mass consistent with BPOP. This imaging evolution is reminiscent of the post-traumatic heterotopic ossification that most radiologists are familiar with in the extremities. In the small bones of the hands and feet, FRP and BPOP have largely been discussed as distinct entities because of their radiographic and microscopic appearances at the time of diagnosis and because of the lack of imaging documentation of FRP evolving into BPOP. Additionally, unlike myositis ossificans or post-traumatic heterotopic ossification in the customary sites where there often is a clear history of antecedent trauma, such a history is not readily found in patients with FRP and BPOP. In our two adult patients, there clearly was no history of trauma, and in the child there was a suggestion that the swelling may have developed 2 weeks after a basketball injury, which is not the usual presentation of an acute traumatic event. It is unclear as to what triggers the striking soft tissue swelling, periosteal reaction and ossification, and it is virtually impossible to prove whether the inciting factors for FRP and BPOP are the same. It would appear, however, that they represent different ends of the spectrum of the same entity, as illustrated in our case 1. It is open to debate whether Fig. 2D represents the far end of FRP or early BPOP. Histological overlap in appearances between FRP, BPOP and reaction to injury has been observed, although the classic microscopic features of FRP and BPOP were not seen in patients with known injury [5]. We tend to agree with the view expressed that the appearances of these entities depends on temporal factors and would add that FRP may be self-limiting or could progress to BPOP [6].

FRP and BPOP favor the proximal and middle phalanges, unlike subungual exostosis which has a predilection for the distal phalanges and is a reactive proliferation that shares some of the histological features found in FRP and BPOP. FRP uncommonly may involve the thumb [8] and cause irregularity of the cortex with erosion [9, 10]. FRP and BPOP have been described in long bones [11, 12].

A significant difference between two large series of FRP and BPOP in the small bones of the hands and feet was the outcome following surgery, there being virtually no recurrences in the patients with FRP and a little over 50% recurrence reported in patients with BPOP [1, 4]. Follow-up of over 1 year in three of our patients shows no evidence of recurrence. In two of our patients, there has been complete resolution of symptoms with normal function. In one of our patients, there still is some residual soft tissue swelling.

Although rare, FRP and BPOP have been relatively well defined over the past two decades. Malignant bone tumors of the hands and feet, on the other hand, although also rare, are entities with which there is a long historical familiarity. Their incidence is well entrenched in the radiology and pathology literature. Despite its newness and relative rarity, FRP is, on a statistical basis, a more common lesion of the small bones of the hands than osteosarcoma and is a diagnosis that should be considered when a patient presents with a large soft tissue mass, an intact phalanx and an aggressive periosteal reaction. The more immediate worry in our view in the prospective evaluation of these patients is osteomyelitis or a soft tissue infection. An early follow-up radiograph within 7–10 days, viewed in conjunction with the clinical findings, ought to clearly separate these entities.

There is no body of information as to the ideal timing for removing these lesions, but it is clear from the literature that most of these lesions have been removed for fear of missing a more sinister pathological disease process and have often recurred. We believe that FRP is a diagnosis that ought to be considered, prospectively, more frequently than it appears to be and that close, careful clinical and imaging follow-up can avoid unwarranted arbitrary antibiotic treatment or hastily performed surgical procedures. In its evolution it may progress to BPOP.

Acknowledgment The authors gratefully acknowledge the assistance of Mrs. Lois Hebel in the preparation of this manuscript.

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