

Available online at www.sciencedirect.com

## **ScienceDirect**



journal homepage: www.elsevier.com/locate/radcr

### **Case Report**

# Focal reactive periostitis ossificans in a long bone: radiologic and pathologic findings \*

# Jennifer L Levy, MD<sup>a</sup>, Katie L Louka, MD<sup>b</sup>, Kumarasen Cooper, MD<sup>b</sup>, Kristy L Weber, MD<sup>c</sup>, Saeed Dianat, MD<sup>a,\*</sup>

<sup>a</sup> Division of Musculoskeletal Radiology, Department of Radiology, University of Pennsylvania, 3400 Spruce Street, Philadelphia, PA, USA 19104

<sup>b</sup> Division of Bone and Soft Tissue Pathology, Department of Pathology, University of Pennsylvania, Philadelphia, PA, USA

<sup>c</sup> Division of Orthopedic Oncology, Department of Orthopedics, University of Pennsylvania, Philadelphia, PA, USA

#### ARTICLE INFO

Article history: Received 21 August 2021 Revised 4 September 2021 Accepted 4 September 2021

Keywords: Florid reactive periostitis ossificans surface lesions reactive

#### ABSTRACT

Florid reactive periostitis ossificans (FRPO) is a benign juxta-cortical lesion of unknown etiology which most commonly occurs in the hands and feet. We report the radiographic, CT, and MR features of a pathologically confirmed FRPO in the distal femur, a location in which only a handful of cases has been reported. A 26-year-old male who presented with distal thigh pain initially underwent radiograph and CT, which illustrated a well-circumscribed, ossified lesion associated with the cortex of the femur without contiguity with the medullary canal. A subsequent MRI demonstrated heterogeneous signal intensity corresponding to the ossified portion of the lesion with a T2 hyperintense cartilaginous cap and surrounding edema. The lesion was surgically excised and pathologic diagnosis of FRPO, a mixture of osteoid, mature bone, cartilage and fibrous tissue, with associated inflammatory cells, was confirmed. Follow up four months after surgery revealed significant improvement in the patient's pain. © 2021 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license

(http://creativecommons.org/licenses/by-nc-nd/4.0/)

#### Introduction

Reactive bone lesions may pose diagnostic challenges, as a history of prior trauma is not always present and the radiographic and histopathological findings can vary depending on the degree of maturity of the lesion and the time since injury. Subperiosteal new bone formation occurs, a process which may be associated with erosion of the osseous cortex causing confusion with an aggressive or malignant process [1-3]. Rarely, the reactive process results in a pedunculated mass which is distinguished from an osteo-

 $<sup>^{*}</sup>$  Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

<sup>\*</sup> Corresponding author.

E-mail address: saeed.dianat@pennmedicine.upenn.edu (S. Dianat).

https://doi.org/10.1016/j.radcr.2021.09.009

<sup>1930-0433/© 2021</sup> The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 – Lateral radiograph of a 26-year-old male patient presenting with pain and swelling demonstrates a well-circumscribed, ossified lesion arising from the cortex of the distal femur which does not demonstrate contiguity with the medullary canal. There is thickening or reactive change of the adjacent cortex (arrow).

chondroma by the lack of cortical and medullary continuity [4].

Florid reactive periostitis ossificans (FRPO) is one such benign reactive bone lesion which is commonly seen in the hands and feet and only rarely reported in the long bones. In this report, we present a 26-year-old man with a history of several months of anterior thigh pain, who is found to have FRPO of the distal femur.

#### **Case Report**

A 26-year-old male without significant past medical history presented with anterolateral distal thigh pain for four months, worsened after doing squats and lower body exercises at the gym. He initially attempted to relieve the pain by resting for a couple of weeks but this did not alleviate his symptoms. On physical examination, significant impingement with flexion and extension was observed. The patient underwent a radiograph of the knee (Fig. 1) which demonstrated a wellcircumscribed, ossified lesion associated with the cortex of the femur without contiguity with the medullary canal. Reactive change of the adjacent cortex was noted. A subsequent CT (Fig. 2) demonstrated an ossified lesion associated with the cortex with peripheral mineralization as well as the adjacent periosteal reaction that was seen by radiograph. An MR demonstrated heterogeneous T2 signal intensity associated with the ossified segment and a T2 hyperintense cartilaginous cap (Fig. 3A and B). Cortical disruption as well as surrounding bone marrow edema were noted. Post-contrast sequences (Fig. 3C) revealed enhancement surrounding the lesion and at the interface of the cartilaginous and ossified portions.

The lesion was surgically resected four months after the initial onset of the patient's pain and two weeks after presentation to our institution. H&E slide of the pathologic specimen (Fig. 4) demonstrated a subperiosteal proliferation of bland but hypercellular and disorganized chondrocytes with reactive myxoid change. At the interface of cartilage and trabecular bone, active endochondral ossification was seen, with woven bone formation in varying degrees of mineralization. Prominent osteoblastic rimming and scattered osteoclastic activity were also present.

In follow up four months after the surgery, after six weeks of physical therapy, the patient reported significant improvement in his pain and range of motion of the knee.



Fig. 2 – Axial (A) and sagittal (B) CT examinations of the knee demonstrate an ossified lesion associated with the cortex with peripheral mineralization (long arrow) as well as the adjacent periosteal reaction that was seen by radiograph (short arrow).





Fig. 3 – MRI axial (A) and sagittal (B) T2 fat-suppressed sequences demonstrate a lesion arising from the anterior cortex of the distal femur composed of a heterogeneous ossified segment (long arrow) with a T2 hyperintense cartilaginous cap (\*). Cortical disruption (short white arrow) is also noted in this case. There is associated bone marrow edema. On axial post-contrast T1 fat-suppressed sequence (C), there is enhancement surrounding the lesion and at the interface of the cartilaginous and ossified portions.

#### Discussion

Florid reactive periostitis ossificans (FRPO) is a benign juxtacortical lesion of unknown etiology. It was initially believed to result from trauma, although such a history is not always elicited [1]. Patients typically present in their third decade with painful swelling and erythema of the affected area [5-8]. Although it predominantly affects the tubular bones of the hands and feet, FRPO is rarely seen in long bones, as in our case [7, 9].

FRPO was first described in 1981 [5]. It is believed to represent part of a continuum of benign reactive juxtacortical lesions, which also includes bizarre parosteal osteochondromatous proliferation (BPOP) and Turret exostosis, since progression from FRPO to BPOP has been reported [8].

Radiologic findings include an ossified lesion arising from the cortex, often with cortical thickening, but which does not demonstrate contiguity with the medullary canal (Fig. 1) [6,10]. While the cortex is usually intact, there are reports of cortical erosion, presumably a result of rapid growth of the lesion [5-8]. This finding was observed in our case. Periosteal reaction, and peripheral mineralization are also sometimes observed (Fig. 2) [4,10]. Soft tissue swelling, typically out of proportion to the extent of periosteal reaction, can be a helpful sign to distinguish it from a malignant tumor or osteomyelitis [8].

MRI demonstrates heterogeneous signal on T2-weighted images, often with associated bone marrow and soft tissue



Fig. 4 – H&E slide at low power (10x) (A) shows a subperiosteal proliferation of chondrocytes (short black arrow) with reactive myxoid change (white arrow). Deeper into the lesion, at the interface of cartilage and trabecular bone, active endochondral ossification (long black arrow) can be seen. At higher power (20x) (B), the superficial portion of the lesion shows bland but hypercellular and disorganized chondrocytes (bracket). Beneath this is endochondral ossification (black arrow) with woven bone formation in varying degrees of mineralization (\*). Prominent osteoblastic rimming (long white arrow) and scattered osteoclastic activity (short black arrow) are present.

edema [7,10]. On post-contrast images, the lesion demonstrates rim enhancement with a non-enhancement of the central portions of the ossified and cartilaginous areas (Fig. 3) [6].

99mTc-MDP bone scan and 18F-FDG PET/CT show increased tracer uptake, findings which may be attributable to increased osteoid production and hypercellular spindle cell proliferation, respectively [9].

Pathologic findings in FRPO include a mixture of osteoid, mature bone, cartilage and fibrous tissue, often with associated inflammatory cells (Fig. 4) [6]. In contrast to malignant lesions, there is no cellular pleomorphism or mitotic figures [4].

The important differential consideration is juxtacortical osteosarcoma. When it occurs in the hands and feet, a very rare location for parosteal osteosarcoma, FRPO can be readily differentiated from osteosarcoma based on location [11]. However, the distinction becomes more challenging in the long bones, as in our case, and pathologic confirmation is often necessary. While the presence of an intact cortex generally favors FRPO over osteosarcoma, this feature is not always present in FRPO and parosteal osteosarcoma can also be seen with an intact cortex [11].

Treatment of FRPO is controversial. Although excision has historically been preferred, local recurrence has been reported [4,5, 9,10]. Therefore, some advocate observation, particularly in the long bone FRPO, which may resolve on its own in 4-8 months with normalization of inflammatory changes and only a residual, painless exostosis [7,10]. Of course, the location of the lesion must be taken into consideration; prominence under the patellar tendon seen in our case would likely not have been amenable to conservative treatment.

In summary, FRPO is a benign juxta-cortical lesion which may exist on a spectrum with BPOP and Turret exostosis. It must be differentiated from osteosarcoma particularly in the long bones, which often requires pathologic confirmation. Historically, excision has been recommended but recently conservative treatment has been advocated in select cases.

#### Patient consent

Informed consent was obtained from the patient for publication.

#### REFERENCES

- Jambhekar NA, Desai SS, Puri A, Agarwal M. Florid reactive periostitis of the hands. Skeletal Radiol 2004;33:663–5.
- [2] Jongeward RH Jr, Martel W, Louis DS, Okoye MI, Walter N. Case report 304. florid reactive periostitis proximal phalange of the left 5th finger. Skeletal Radiol 1985;13(2):169–73.
- [3] Callahan DJ, Walter NE, Okoye MI. Florid reactive periostitis of the proximal phalanx. Case report. J Bone Joint Surg Am 1985;67(6):968–70.
- [4] Porter AR, Tristan TA, Rudy FR, Eshbach TB. Florid reactive periostitis of the phalanges. Am J Roentgenol 1985;144(3):617–18.
- [5] Spjut HJ, Dorfman HD. Florid reactive periostitis of the tubular bones of the hands and feet: a benign lesion which may simulate osteosarcoma. Am J Surg Pathol. 1981;5:423–33.
- [6] Azorin D, Lopez-Pino MA, Gonzalez-Mediero I, Epeldegui T, Lopez-Barea F. Long bone florid reactive periostitis ossificans: a case in the distal femur mimicking osteosarcoma. J Pediatr Orthop B 2008;17:301–5.
- [7] Brien EW, Zahiri CA, Mirra JM. Florid reactive periositis ossificans of the proximal aspect of the tibia: a lesion that must be distinguished from osteosarcoma. J Bone Joint Surg Am 1999;81:1002–7.
- [8] Sundaram M, Wang L, Rotman M, Howard R, Saboeiro AP. Florid reactive periostitis and bizarre parosteal

osteochondromatous proliferation: pre-biopsy imaging evolution, treatment and outcome. Skeletal Radiol 2001;30:192–8.

- [9] Byun BH, Koh JS, Yoo JY, Lim SM, Kong CB. (99m)Tc-MDP- and (18F)-FDG-avid florid reactive periositis ossificans mimicking recurrent osteosarcoma. Clinical Nuclear Medicine 2013;38(6):482–3.
- [10] Jamshidi K, Givehchian B, Mirzaei A. Florid reactive periostitis of the long bone: a case series of seven patients. J Orthop Sci 2017;22(3):560–5.
- [11] Stark HH, Jones FE, Jernstrom P. Parosteal osteogenic sarcoma of a metacarpal bone: a case report. J Bone Joint Surg 1971;53:147–53.