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Florid Reactive Periostitis of the Tibia: Case Report

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Abstract

Florid reactive periostitis is a rare bone proliferation with a pronounced periosteal reaction. These lesions are rarely described in long bones since they generally appear in the tubular bones of the extremities. The etiology is unknown, although it is related in several cases to a traumatic history. It shares imaging and histological similarities with other bone lesions such as Bizarre Parosteal Osteochondromatous Proliferation (BPOP), subungual exostosis, osteomyelitis, myositis ossificans, and malignant surface tumors of bone and cartilage which include conventional, periosteal and parosteal osteosarcoma. Regarding treatment when presenting early, this process can be treated conservatively with rest and non-steroidal anti-inflammatory medication. When presenting late, with an aggressive nature and/or with recurrence, wide local resection is considered the treatment of choice. Differential diagnosis is essential to avoid unnecessary aggressive treatments. We present a case of a florid reactive periostitis of a middle third tibia, with progressive growth without previous trauma or infectious process. The diagnosis was not suspected by imaging tests. The histological findings of a parosteal lesion with benign characteristics raise the possibility of florid reactive periostitis, having ruled out chronic osteomyelitis due to the negative microbiological studies. After the intervention, the pain disappeared and one year after the surgery there were no recurrences of the lesion.

Introduction

Florid Reactive Periostitis (FRP) is a rare benign periosteal lesion, which mostly involves the tubular bones of the extremities. FRP of the long bone is especially rare. We present a case of a florid reactive periostitis of a middle third tibia.

Case Presentation

A 36-year-old female patient presented with a swollen and painful lesion in the middle third of the right tibia with progressive growth without previous trauma or infectious process in the tibia. Characteristically predominant nocturnal pain. Conventional radiographs revealed a bone tumor with periosteal reaction with involvement of adjacent soft tissues (Figure 1). Due to the intense pain, the patient went to the emergency room and a deep vein thrombosis was ruled out by Doppler ultrasound showing an area of cortical bone irregularity with periosteal reaction. The MRI showed a lesion in the middle third of the diaphysis of the right tibia with Codmann's triangle and associated medullary edema (Figure 2). The CT scan showed a lesion of aggressive characteristics with periosteal reaction in onion layers and with prominent speculation that affects more than 90% of the transverse section of the tibia and a craniocaudal extension of 6 cm (Figure 3). With these findings, extension study and bone biopsies were carried out by tru-cut for radiodiagnosis. In the histological analysis, only soft tissues with myxoid edematous changes were observed. Given the hardness of the bone lesion, it was not possible to take a sample of it percutaneously. A second incisional biopsy was performed with these results: Very compact cortical and medullary bone with portions of mature lamellar bone with ossification and mineralization. The arrangement of some trabecula was parallel and others more irregularly distributed. It showed a florid crown of osteoblasts in some areas without atypia with very occasional mitotic figures. Given the radiological data of the lesion on the cortical-dependent surface, FISH of the MDM2 gene was performed, not observing its amplification. The sample sent as periosteum showed chronic inflammatory signs and vascular proliferation. Samples are sent to microbiology, all being negative. The study of the complete piece was recommended, as it may be a florid ossifying reactive periostitis. Surgical intervention was carried out, performing an extended resection, leaving the lateral hemicortical bone of the tibia, and a cancellous structural graft was provided and stabilized with a plate (Figure 4, 5). Several samples are sent to pathological anatomy and microbiology (Figure 6). Histological results again showed a proliferative bone lesion with trabecula that are arranged perpendicular to the cortical bone, the

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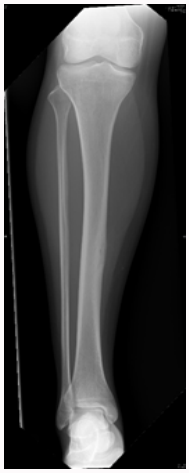

Figure 1: Antero-posterior X-ray revealed a bone tumor with periosteal reaction with involvement of adjacent soft tissues.

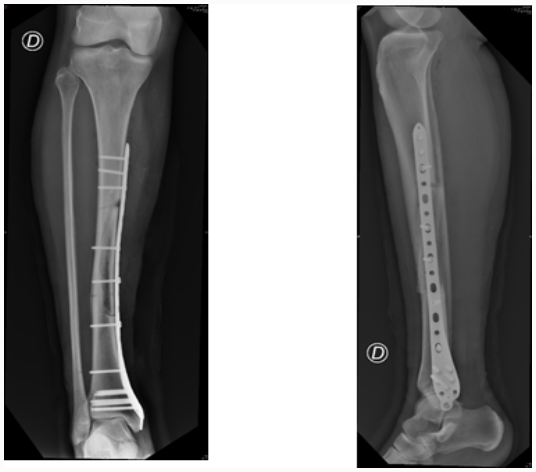

Figure 4: Postoperative X-Ray show the cancellous structural graft at the excision´s site of the bone lesion fixed with a plate.

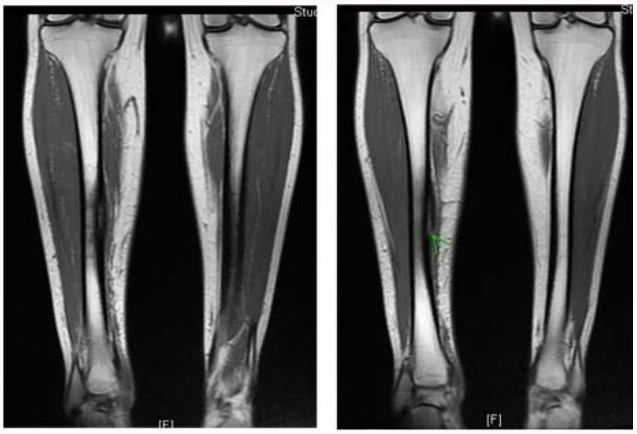

Figure 2: MRI shows a lesion in the middle third of the diaphysis of the right tibia with Codmann's triangle and associated medullary edema.

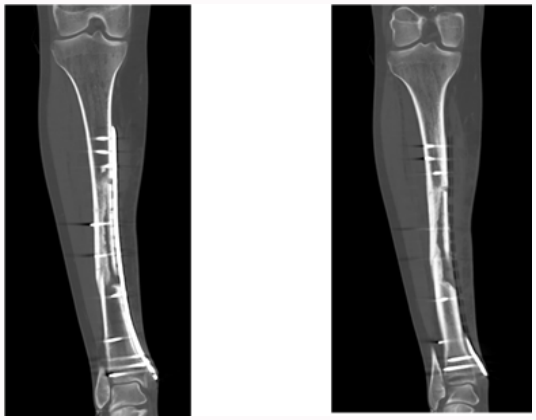

Figure 5: Postoperative CT images.

bone had a mature appearance with more densely sclerosed areas and empty lacunae of osteocytic necrosis and phenomena of reactive bone formation could be glimpsed. It was a sclerosing lesion. In continuity and towards the surface, periosteal reaction with neoformation and vascular proliferation as well as fibroblasts was observed. As no areas of chondroid differentiation or frank atypia were observed (Figure 7), the diagnosis of periosteal osteosarcoma was ruled out. A new FISH of the MDM2 gene was performed, not observing amplification, so parosteal osteosarcoma was ruled out. Therefore, the findings of a parosteal lesion with benign characteristics raise the possibility of florid reactive periostitis, having ruled out chronic osteomyelitis due to the negative microbiological studies of both surgeries. After the intervention, the pain disappeared and one year after the surgery there were no recurrences of the lesion.

Discussion

Florid reactive periostitis is a benign bone lesion characterized by

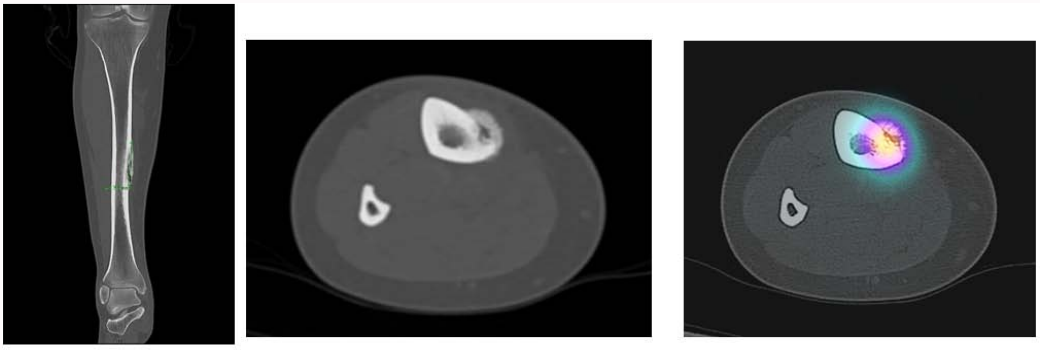

Figure 3: CT shows a lesion of aggressive characteristics with periosteal reaction in onion layers and with prominent speculation that affects more than 90% of the transverse section of the tibia and a craniocaudal extension of 6 cm. Gammagraphy shows increased uptake at the bone lesion.

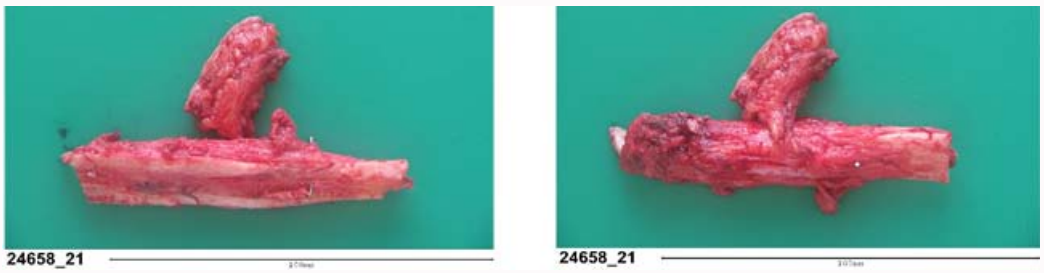

Figure 6: Volar and dorsal view of the surgical piece after tumor excision and biopsy path of about 10 cm in length. Macroscopically, cortical increases were only observed in the tumor area.

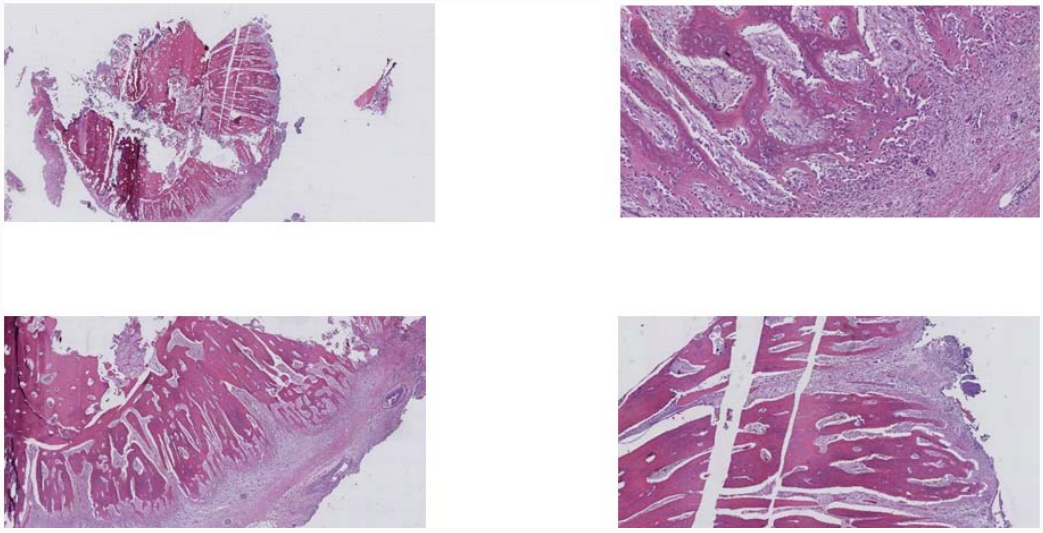

Figure 7: Pathologycal analysis with hematoxylin-eosin technique describes a bone fragment that shows in the surface of the cortical bone proliferative lesion whose trabeculae are arranged perpendicular to the cortex. At the base of the lesion, there is mature-looking bone with cement lines and areas more densely sclerosed. In continuity and towards the Surface, is observed periosteal reaction with new reactive bone formation, with abundant polygonal osteoblasts and some mitotic figures. Focally there is infiltrate inflammation between the bone components.

an aggressive periosteal reaction and soft-tissue inflammation [1,2]. Usually affects the digits of the hand and less frequently the feet. It is characterized by pain and tight swelling restricting digital motion and is not usually related to previous trauma. The imaging and histologic features, show an overlap with other bone lesions including BPOP, subungual exostosis, osteomyelitis, myositis ossificans, and malignant surface tumors of bone and cartilage which include, conventional, periosteal and parosteal osteosarcoma. From a histological point of view, cartilage is absent in FRP and periosteal reaction is absent in BPOP [3]. On BPOP the exophytic bone mass has a characteristic dark blue tinctorial quality, especially at the interface with the cartilage [4]. Another benign lesion that occurs in relation to the long bones (and shows overlapping radiologic and histologic findings with reactive periostitis) is myositis ossificans traumatic [5]. Peripheral ossification is the characteristic feature noted on CT [5,6]. Histologically there are three phases of myositis ossificans: acute (fibroblastic zone), subacute (osteoblastic zone), and late (calcified zone). Detection of this zonal phenomenon on histology is diagnostic of myositis ossificans [5,6]. Another benign lesion that occurs in relation to the long bones (and shows overlapping radiologic and histologic findings with reactive periostitis) is myositis ossificans traumatic [5]. Peripheral ossification is the characteristic feature noted on CT [5,6]. Histologically there are three phases of myositis ossificans: acute (fibroblastic zone), subacute (osteoblastic zone), and late (calcified zone). Detection of this zonal phenomenon on histology is diagnostic of myositis ossificans [5,6]. Chronic osteomyelitis may also be in the differential diagnosis. This diagnosis is made radiologically by the presence of an osteolytic center with a ring of sclerosis on conventional imaging [7]. Additionally, a culture of the biopsy tissue is needed to support the diagnosis and identify the specific pathogen. Histology often only shows sclerotic bone with chronic inflammatory cells [8]. Furthermore, subungual exostoses can also mimic florid reactive periostitis. There is a reproducible translocation [t(x; 6) (q13; q22)] associated with this diagnosis and thus, it may be considered a true neoplasm. Lastly, malignant osteoblastic tumors including parosteal, periosteal, and conventional osteosarcoma are considered in the differential diagnosis. Parosteal osteosarcoma is the most frequently occurring osteosarcoma and thus it is crucial to diagnostically separate this entity from reactive periostitis. Histologically, it exhibits an extensive boney matrix with a hypocellular stroma and mild to minimal fibroblastic cellular atypia. Radiologically, it takes the appearance of a firm, lobulated “cauliflower-like”, lesion encircling the bone. A thin radiolucent line delineating the tumor from the cortex, known as the “string sign”, is seen radiologically in 30% of cases conventional osteosarcoma. Histologically, these tumor cells are very pleomorphic with numerous atypical mitoses that are entrapped in the osteoid matrix. Regarding treatment when presenting early, this process can be treated conservatively with rest and non-steroidal anti-inflammatory medication [9]. When presenting late, with an aggressive nature and/or with recurrence, wide local resection is

considered the treatment of choice.

Conclusion

Florid Reactive Periostitis Ossificans (RPO) is a rare benign entity that in less than 100 cases has been described in the literature with far fewer outside the bones of the hand, feet, fingers, and toes. Because the imaging and histologic features show an overlap with other bone lesions and malignant conflict tumors of bone and cartilage a careful assessment of clinical history, radiology, and pathology help reach an accurate diagnosis. Although rare, this entity should be considered in the differential diagnosis of any osteogenic growth in long bones.

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