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Co-existence of Non-ossifying Fibroma and Osteoid Osteoma in one Tibia: a Case Report

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Abstract

Presentation of two independent tumors in one case is rare; coincidence of two different bone tumors in a single bone and limb is extremely rare. Here we reported on a male adolescent case with co-existence of NOF and an osteoid osteoma in ipsilateral tibia which was presented with left leg pain and swelling; both tumoral lesions were excised and the patient became symptom free. This case study highlights the importance of careful clinical and radiological investigation for more pathologies in spite of finding a single tumor; and encourages further research on the possible genetic links between these two kinds of tumors.

Key Words: Bone neoplasms, Metaphyseal fibrous defect, Non-ossifying fibroma, Osteoid osteoma, Tibia.

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1- INTRODUCTION

Non-ossifying fibroma (NOF), also termed as metaphyseal fibrous defect, is a rather common fibroblastic tumor lesion of long bones, arising in 20-50% of children and adolescents (1-3).The lesion is often asymptomatic and detected on radiographic images (2, 4).

Most NOFs require no therapeutic intervention; though, curettage and bone packing might be needed for symptomatic treatment; and large lesions involve more than 50% of the diameter of the bone because of a potential chance of pathologic fracture. Recurrence after curettage is usually rare (5-7).

Osteoid osteoma, as a benign osteoblastic lesion, was first introduced in 1935. It is typically seen in adolescents and young adults in lower extremities; so, the patients usually present with pain and limping (8). Conventional X-rays beside clinical symptoms are sufficient in the diagnosis but the biopsy is rarely required to confirm the diagnosis. CT scan is the modality of choice for diagnosis; a central radiolucent area (nidus) bounded by a zone of cortical thickening would be noted (8, 9). MRI is only limited to the unusual and difficult diagnoses and CT-contraindicated cases (7). The microscopic pathology manifestation consists of asclerotic rim of fibrovascular stroma with immature bony trabeculae surrounded by dominant osteoblasts. Such an appearance is similar to that of an osteoblastoma but the osteoblastomas are larger. There is no nuclear atypia nor any aggressive trait; the tumor presents classically in two thirds of the patients (5, 10, 11). The Juxtacortical and subperiosteal location of the tumor would make the diagnosis difficult. Osteoid osteoma, sometimes, arises in the subperiosteal region initially and becomes cortical or intramedullary later in its course (12, 13).

Treatment strategies include conservative medical therapy, percutaneous radiofrequency ablation, and open surgeries. Surgical management consists of the entire nidus removal via tumor curettage or en-bloc resection; the latter may have a low rate of recurrence (13, 14).

The concurrence of a NOF with a primary bone tumor is exceptionally rare, with limited cases reported previously (8, 10, 15-19).

Here we reported a case with NOF of distal tibia coinciding with the same tibia tumor of osteoid osteoma in his proximal.

2- CASE PRESENTATION

A 16-year-old white boy was presented to our orthopedic clinic at Akhtar Hospital because of left proximal leg swelling and pain following a direct trauma. On physical examination, his proximal tibia was tender with no erythema; the knee and ankle's range of motion were within the normal limits. He was not febrile and had no recent significant weight loss. The vital signs were normal and he had no remarkable past medical history.

Plain radiographies of the affected limb showed proximal tibia juxtacortical opaque sclerotic lesion with soft tissue swelling. Besides, an incidental finding of the same distal tibia metaphysical lesion having multilocular appearance and sclerotic scalloped borders, with cortical erosion of the cortex radiologically in favor of NOF. CT scan of the proximal tibia showed a nidus lesion suggesting Osteoid Osteoma (**Fig. 1**).

Routine laboratory tests were unremarkable except for a mild elevated serum alkaline phosphatase but without proteinuria (**Table 1**).

The patient was scheduled for open excision biopsy; the biopsies of proximal and distal tibia lesions were conducted in the operating room under anesthesia and

tourniquet; the pathology reported osteoid osteoma and NOF for the corresponding lesions (**Figure2** A, B). In addition, the emptied site of the lesions was filled with ipsilateral iliac bone autograft and fixed with an anatomical plate (**Figure3**, A and B). The patient was allowed to full weight bearing after 8 weeks post-operatively.

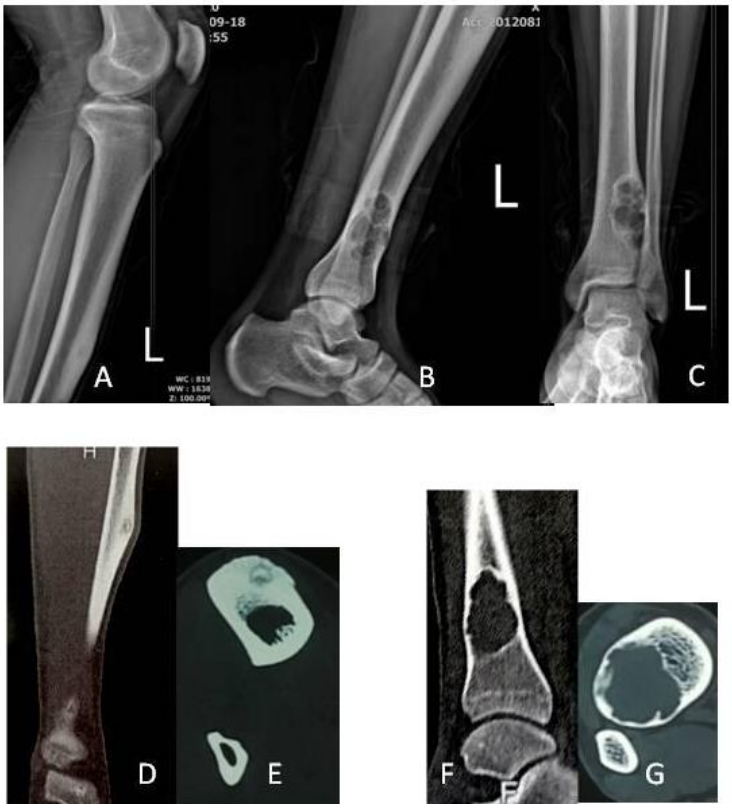

Fig. 1: Preoperative X-rays and CT scans illustrating the characteristic lucent nidus within the surrounding sclerotic reactive bone in proximal diaphysis of tibia characterizing an Osteoid Osteoma (A lateral radiograph, D, E Sagittal and axial CT scan cuts), and distal well defined eccentric cystic multiloculated lucent lesions with a sclerotic rim in the metaphysic of the ipsilateral tibia (B,Anteroposterior and lateral radiographs, F,G Sagittal and axial CT scan cuts).

Table-1: The patient's laboratory blood test results

Test	Result
Red blood cells	4.1 trillion cells/L
White blood cells	4.8 billion cells/L
Hemoglobin	16.8 grams/dL
Hematocrit	38.3 percent
Platelet count	141 billion/L
Erythrocyte sedimentation rate	2 mm/hr
c-reactive protein	5 mg/L
Urea	45 mg/dL
Creatinine	1.2 mg/dL
Alkaline phosphatase	157 IU/L
Lactate dehydrogenase	285 U/L
Calcium	10.1 mg/dL
Phosphorus	3.9 mg/dL

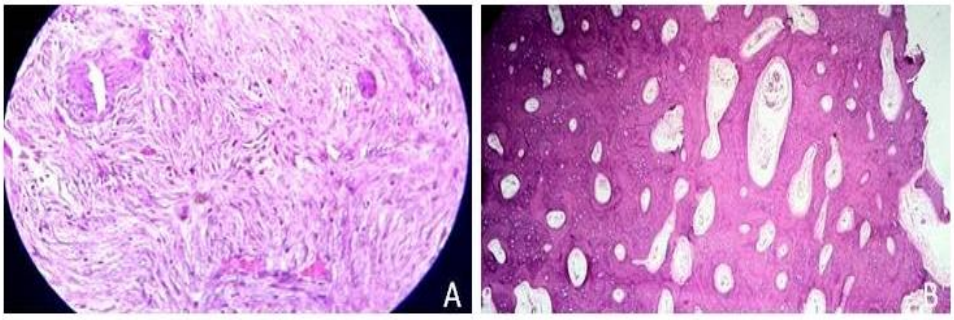

Fig. 2: Distal lesion microscopic appearance characterizing NOF with storiform fibroblasts, scattered benign giant cells, foamy histiocytes, and hemosiderin besides mitotic figures (A). Proximal lesion microscopic view of osteoid osteoma showing small, circumscribed anastomosing, immature trabeculae rimmed by osteoblasts and osteoclasts plus loose fibrovascular tissue (B).

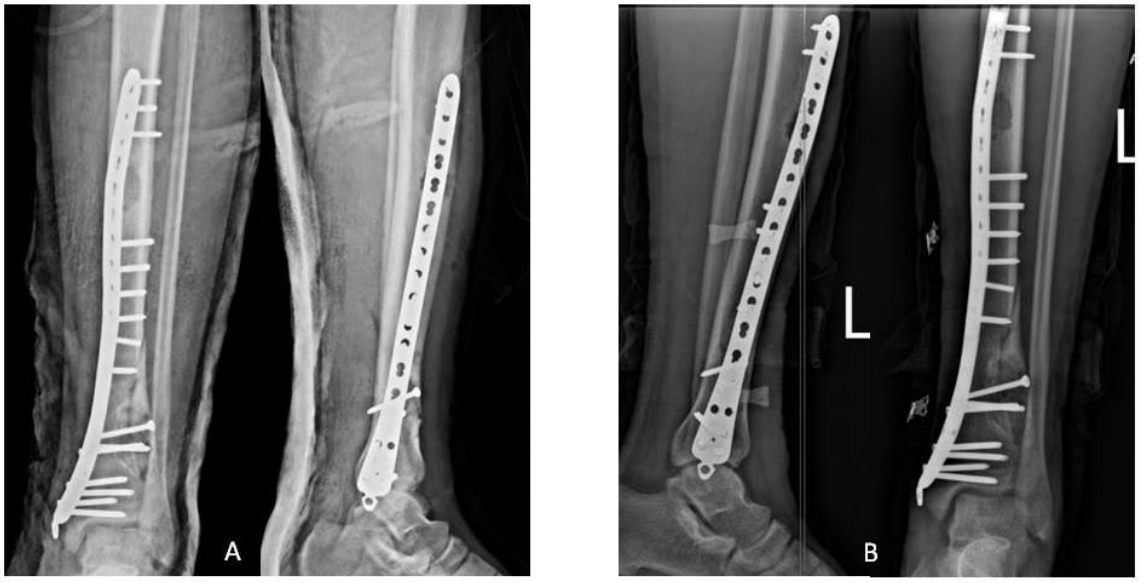

Fig. 3: Immediate Postoperative radiography (A), 3-month postoperative follow-up X-ray (B)

3- DISCUSSION

The concurrence of benign primary bone tumors with NOFs has been infrequently noted; coexistence of osteochondroma of the tibia and a non-ossifying fibroma of the contralateral tibia (20) or osteochondroma of femur and NOF in tibia (21), and osteoid osteoma of proximal femur and NOF in distal femur (22) could be examples. In a previous report it was mentioned that bone fibroma lesions seem to be somehow associated to the adamantinoma of the long bones, and lesions similar to fibrous dysplasia are associated with adamantinoma of the long bones which are in fact ossifying fibroma (23).

Also, an important differential diagnosis for the distal lesion of our case could be giant cell tumor (GCT). However, GCTs are located exclusively in the epiphyseal region, usually, in long bones and consist of a larger number of giant cells in the microscopic analysis (24).

Our case seems to be comparable to those very limited number of previously reported cases mentioned above; radiologic and histopathologic evidence in previous cases

defends the conclusion that they characterize co-existence of two independent tumoral lesions. In our current case, the radiologic and histopathologic evaluation also evidently illustrated two distinct lesions, one of which showed features of a NOF, and the other, represented the features of an osteoid osteoma.

4- CONCLUSION

Considering the previously published case reports, the coexistence of NOF with another unique bone tumor -osteoid osteoma- in the same limb and bone is extremely rare.

This report could be helpful evidence for further Investigations on the possible common genetic origins or mutual associations which cause the arising of two coincided tumoral lesions. Also our case recalls the importance of careful clinical and radiological investigations for more pathologies in spite of finding a single tumor lesion.

5- ACKNOWLEDGMENTS

The authors wish to thank the patient and his family for their compliance. They are also grateful to the department of pathology, Shahid Beheshti University of Medical Sciences, Tehran, Iran for interpreting the histopathology data.

6- ETHICAL CONSIDERATIONS

Informed consent and consent to publish was obtained from our patient and his family whose archived data was reported in the study; the personal data of the patient was kept confidential. The research was supervised by the Ethics Board, Deputy of Research, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

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