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Case Report

Secondary aneurysmal bone cyst with benign fibro-osseous lesions: Case report

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ABSTRACT

Introduction: Aneurysmal bone cyst is a controversial osteolytic benign expansive lesions which occur more frequently in the metaphysis of long bones and spine. They are classified as primary or secondary lesions depending on the presence or absence of an associated bone pathology.
Case presentation: An uncommon case has proven histological of benign fibro-osseous lesions (fibrous dysplasia and juvenile psammomatoid ossifying fibroma) with secondary aneurysmal bone cyst formation inferior and lateral to the knee for a 7-year-old female patient. The lesion was surgically removed and the patient was followed up for 6 months with excellent results without any complications.
Discussion: There are many cases that have previously documented a bone aneurysm cyst, but what distinguishes this case is that it is a secondary type and its association with a benign bony fibrosis.
Conclusion: The combination of the two lesions constitutes a unique and rare case that can add to the medical literature within the orthopedic department.

1. Introduction

The aneurysmal bone cyst is a benign tumor like lesion affects the skeleton consisting of blood-filled spaces of variable size separated by connective tissue septal containing trabeculae or osteoid tissue and osteoclast giant cells. It can be seen in any bone of the body but it tends to grow most on the epiphysis of the long bones (Especially the thigh bone and the larger shin bone …). 80% of the patients are affected during their second decade. It is rare to be affected with the lesion before the age of 5 years old, and it is more common in males than females in a ratio of 2 to 1 and It is often isolated, unlike this case, which is accompanied a benign osseous fibrosis [1,2]. In diagnosing the secondary aneurysmal bone cyst with fibro-osseous lesion, Radiographic findings consist of a central or subperiosteal lesion that appears cystic or lytic intramedullary, expansile, and well-defined lesion in the diaphysis or metaphysis.But confirmation of the diagnosis mainly depends on the results of the histopathology of the biopsy taken from the lesion. Since these two lesions are benign, surgical removal of the entire lesion is the best option for treatment with follow-up to investigate the presence of complications in addition to monitoring the movement of the patient.

This case report has been reported in line with the SCARE criteria [3].

1.1. Case presentation

A 7_year_old girl was admitted to University Aleppo Hospital with complaint of swelling inferior and lateral of the knee. She complained initially of a painless swelling that lasted for two months. Then, age complained of a new onset mild to moderate pain that accompanied the swelling which was consistent during effort. The patient had no history of trauma, chronic diseases or surgery. The study of anterioposterior and lateral knee X-ray revealed osteolytic prominent lesion that occupied the entire width of the fibula in the subepiphyseal region (metaphysis), as it did not pass the epiphysis line, and did not show any periosteal reaction (Fig. 1). There was no extension of the lesion to the adjacent soft tissue or damage to the peroneal nerve passing through the anatomic area of

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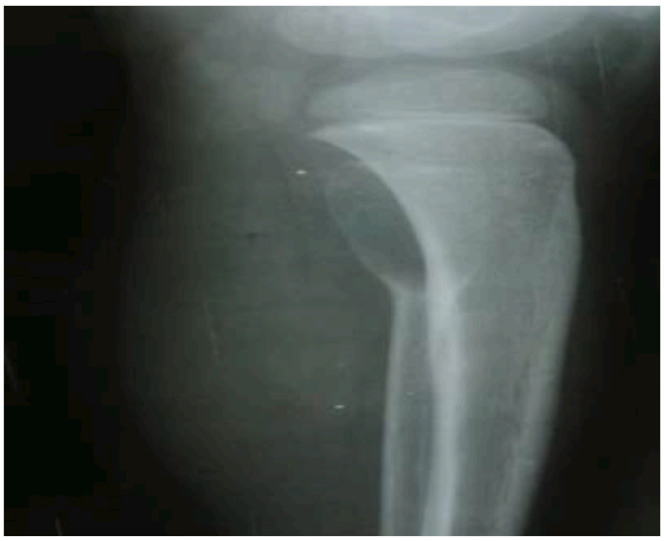

Fig. 1. Lateral knee x_ray that was performed in the first visitvisit, which shows osteolytic lesion.

the lesion. It was scheduled to perform Curettage and grafting type of surgery, but the parents were careless to the importance of the surgery and prevented it. No clinical examination was described for the patient during the first visit. The study of anterioposterior and lateral knee X-ray was repeated, which showed an increase in the size of the lesion without cross the epiphysis line, but there was a new onset periosteal reaction in the lower part of it (a healing fracture with a tumoral periosteal reaction)(Fig. 2). Magnetic Resonance Imaging was requested, but it was not performed by the patient's parent due to a financial problem.

Segmental excision was performed for a suspicious lesion above the fibula in which the entire tumor was excised with the surrounding periosteum and the attachments of the tendons and ligaments, and with complete isolation of the peroneal nerve and preservation of the vascular bundle medial to the lesion (Fig. 3). A drainage tube was

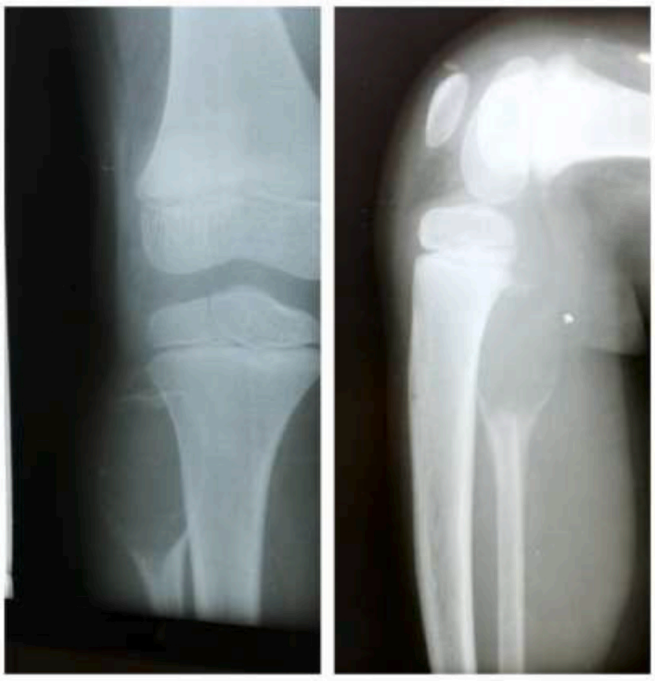

Fig. 2. Anterioposterior and lateral knee x_ray that was performed in the second visit.

Fig. 3. The picture that shows the isolation of fibular nerve during the surgery.

inserted at surgical site. Fortunately, there was no leakage and T was removed after 24 hours. No blood transfusion was needed and the patient state did not require to be admitted into the intensive care unit. The patient was discharged from the hospital 3 days after the operation, during which we used intravenous paracetamol and morphine to relieve the pain and intravenous metronidazole during the operation to avoid the occurrence of postoperative wound infections. We sent the excisional biopsy to the pathologic laboratory and the result confirmed the diagnosis of Benign Fibroosseous tumor with secondary Aneurysmal bone cyst. The gross description showed a fragmented soft and bony pieces. According to the microscopic examination, the sections showed a mass composed of fibrovascular connective tissue infiltrated by large number of osteoclast like giant cells and hemosiderin macrophages, surrounding bony trabeculae (Fig. 4). There were not any necrosis, signs of atypical mitosis or malignancy, but the tumor had extended to the surrounding muscular tissue and cartilage disc. We did an anterioposterior and lateral knee X-ray after surgery(Fig. 5). The integrity of the peroneal nerve was confirmed by clinical examination after surgery

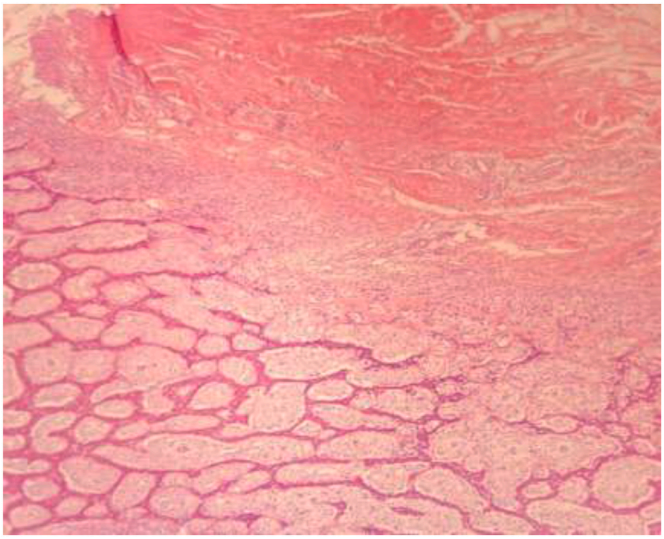

Fig. 4. The histological changes shows benign fibroosseous tumor with secondry aneurysmal bone cyst.

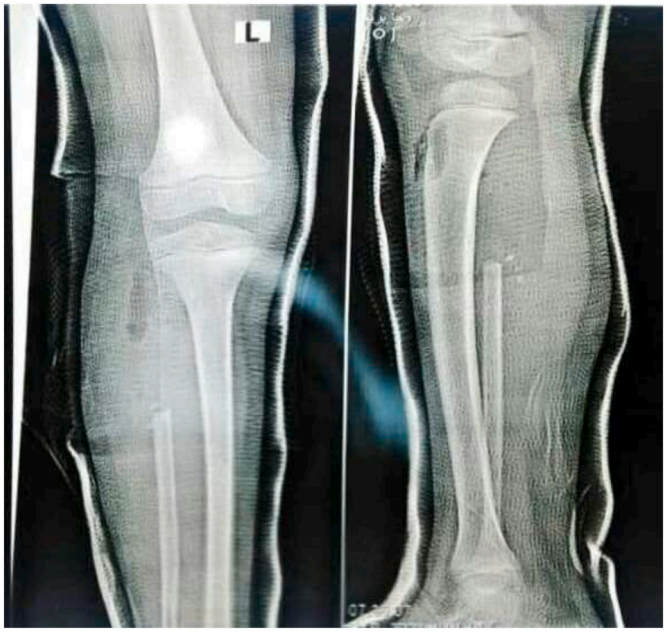

Fig. 5. Anterioposterior and lateral knee x_ray performed after surgery.

and the patient was followed up for 6 months without noticing any sign of complication of the surgery or any nerve damage. Currently, the patient is walking normally without any difficulties.

2. Discussion

The report of aneurysmal bone cyst associated with fibrous dysplasia is an extremely rare. Aneurysmal bone cysts are rare, benign primary bone tumors that can be fast-growing and locally destructive [4] and it is an expanding osteolytic lesion superimposed on an existing pathological process of the bone [1]. Patients with an aneurysmal bone cyst often present with pain in the affected area along with neurological deficits. Fibrous dysplasia is a benign skeletal disorder, first described by Lichtenstein, in which abnormal development of fibroblast replaces medullary bone with fibrocellular tissue [5,6]. ABC appears on plain X-rays as an ovoid lesion with varying degrees of diploic expansion or cortical thinning [1].The characteristic radiographic appearance of osteofibrous dysplasia is eccentric, intracortical, and osteolytic. Variable expansion of the external cortical surface, with sclerosis of the internal cortical surface.

Frequently, a multilocular lesion gives rise to a bubbled appearance. The size of the lesion is variable. Usually, it affects the diaphysis, though metaphyseal encroachment has been reported.On histopathology, Fibrous dysplasia with aneurysm bone cyst have two components. Fibrous dysplasia component is irregular bony trabeculae with varying number of fibroblasts. ABC component is blood-filled cavernous space surrounded by multinucleated giant cells [7]. According to the literature, the treatment of choice for Fibrous dysplasia with aneurysm bone cyst is resection. Also, close follow-up to the resected lesion is recommended. Our patient was a 7-year-old female and the lesion was located in the right fibula bone. Clinically, she was presented with Pain and swelling inferior and lateral to the knee. Radiographic examination reveals an expansile well-circumscribed radiolucent or mixed radiolucent/radiopaque lesion surrounded by a thick bony wall. Histopathology revealed vascular tissue infiltrated by osteoclasts and hemosiderin macrophages in the form of a bony aneurysm cyst surrounded by bony tarabeculae. The cystic nature of the presented lesion, which was also verified intraoperatively, may be attributed to the presence of an associated aneurysmal bone cyst. To our knowledge, this is the 30 s case of a fibro-osseous lesion l with secondary aneurysmal bone cyst to be reported in the literature.

3. Conclusion

The coexistence of benign fibro-osseous lesions and aneurysmal bone cysts is a reality. This rare combination requires accurate radiologicall and histological diagnosis to be treated as a benign tumor that can be treated surgically without chemotherapy or radiotherapy because it is not related to malignancy tumor,which it will add a new knowledge to orthopedic doctors.

Please state any conflicts of interest

All authors declare no conflict of interest.

Registration of research studies

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Author statement

Sarya Swed: contributed in study concept and design, data collection, and writing the paper. Mahmood Islam Kremesh:contributed in writing the paper. Lamees Alshareef: contributed in writing the paper. Jamal Katnaji: contributed in reviewing the paper. Waleed Abd: contributed in reviewing the paper. Kusay Ayoub:contributed in reviewing the paper.

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Consent

Written informed consent was obtained from the patient's parent for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Sarya Swed.

Declaration of competing interest

All authors declare no conflict of interest.

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