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Case Report

A rare co-existence of fibrous dysplasia and aneurysmal bone cyst in the mandible: A case report

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Abstract

Fibrous dysplasia (FD) and aneurysmal bone cyst (ABC) are two different entity, with different pathophysiological mechanisms. FD is a benign fibro-osseous lesion characterized by a fibrous replacement of the medullary component of bone. The second lesion, ABC, is an expansive osteolytic lesion consisting of blood-filled spaces. The coexisting and association of these two disorder is unusual.

We report a case of a 12 year old patient presenting with both FD and ABC in the left mandibular area, and managed surgically in the oral surgery department of the dental consultation and treatment of Casablanca.

Keywords: Fibrous dysplasia, Aneurysmal bone cyst, Management.

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1. Introduction

Fibrous dysplasia (FD), firstly described in 1938 by Jaffe and Lichtenstein,[1-3] is an uncommon slowly progressive, self-limiting, and non-malignant fibro-osseous disorder,[1,4] in which the normal bone marrow is replaced by an abnormal proliferation of a new fibrous connective tissue.[5,6]

There are 2 clinical forms of fibrous dysplasia: monostotic FD that involves only a sign bone and polyostotic that affects different bones.[1,6]

Aneurysmal bone cyst (ABC), on the other hand, was first described by Van Arsdale in 1893 as an ossifying hamartoma, but renamed later by Jaffe and Lichtenstein.[4] It is a rare benign, expansive, and locally destructive lesion, characterized by the presence of blood filled cavities, separated by septa of trabecular bone or fibrous tissue that contain osteoid material and multinucleated giant cells.[4,7-9] It is considered as a pseudocyst due to the absence of real epithelial lining.[4]

The simultaneous occurrence of these two disorders in the same anatomical region is exceedingly rare.

The objective of this article is to present a rare case of 12 year-old male patient diagnosed with both FD and ABC in the pre-angular mandibular area and to discuss the clinical and radiographic features, as well as the therapeutic management of theses lesions.

2. Observation

A 12 year-old male patient with no relevant medical history, presented to the oral surgery unit of the consultation and treatment center of Casablanca, reporting a slow growing swelling in the left mandibular area, developing over the past two months.

The extraoral examination showed a firm, non-painful swelling in the left pre angular mandibular area (**Figure 1**).

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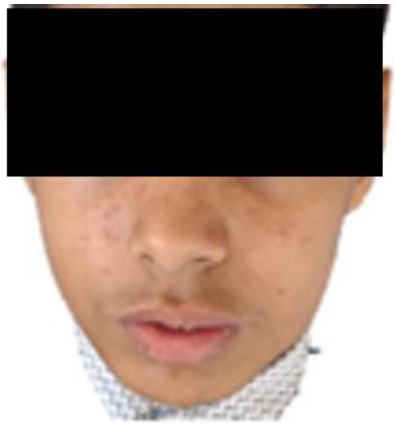

Figure 1: Extra-oral sweling in the left area

The intraoral examination revealed a vestibular swelling opposite the tooth 36 and 37. It was firm on palpation, painless and covered with normal appearing mucosa (**Figure 2**). The pulp vitality of teeth 36 and 37 is maintained.

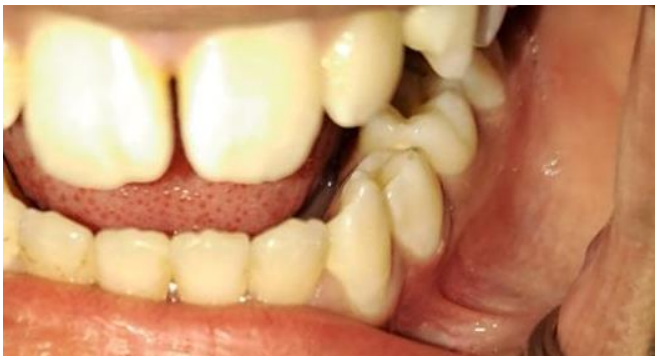

Figure 2: Intra-oral image showing a non-limiting vestibular swelling

The panoramic radiograph showed a well-defined radiolucent lesion, extending from the mesial surface of the tooth 36 to the distal surface of the tooth germ of 38 (**Figure 3**).

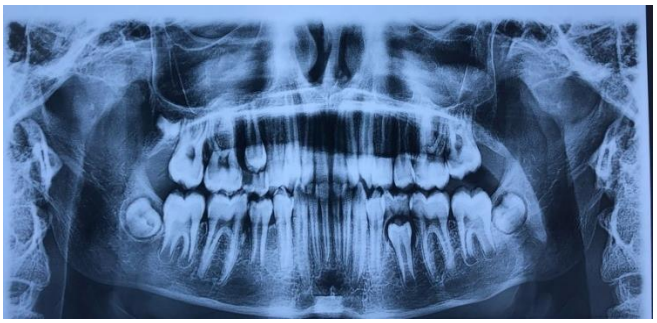

Figure 3: A panoramic radiograph showing a well-defined radiolucent lesion involving the apices of the tooth 36 and the immature tooth 37

Cone beam computed tmography (CBCT) confirmed the presence of a well-defined homogeneous radiolucent lesion located apically and buccally in regard to the teeth, with expansion and thinning of the external cortical bone.

The lesion was in close proximity to their roots, and the inferior alveolar nerve is displaced lingually to the lesion (**Figure 4**).

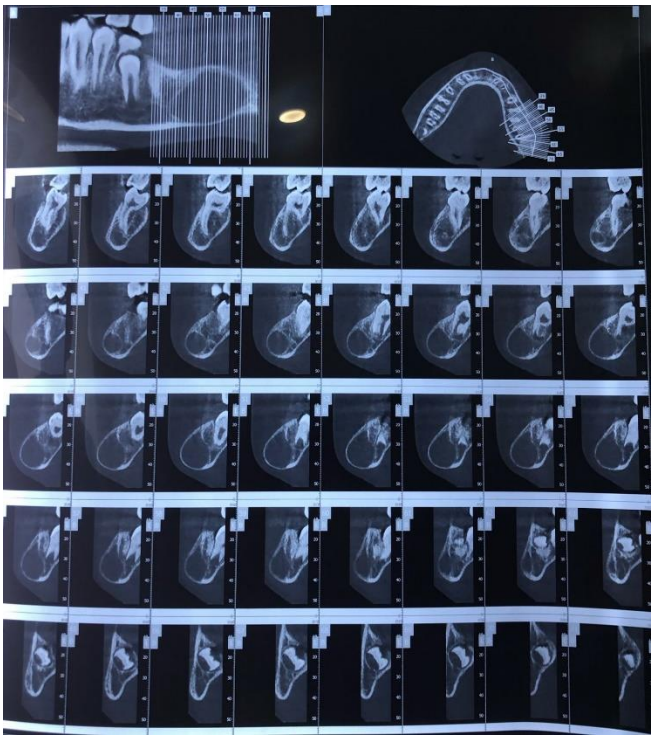

Figure 4: CBCT showing the well-defined radiolucent lesion with thinning of the buccal cortical bone

Based on the clinical and radiographic findings, the differential diagnoses included: ameloblastoma, solitary bone cyst, and odontogenic ketarocyst.

An enucleation and curettage following a bone trepanation were conducted for the treatment (**Figure 5**). Postoperative care included the prescription of an antibiotic (amoxicillin), a non-steroidal anti-inflammatory drug, an analgesic and an antiseptic mouthwash.

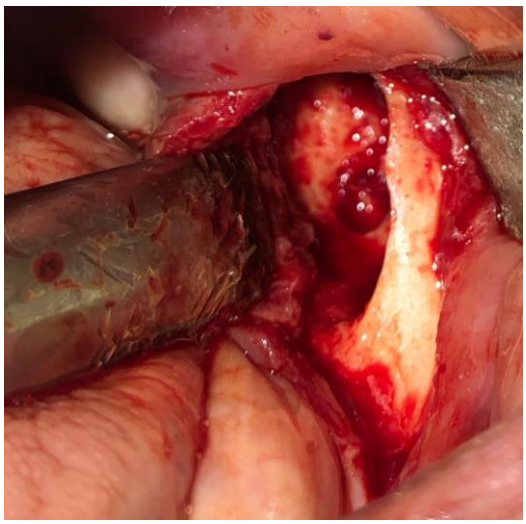

Figure 5: Intraoperative photo after enucleation

During surgery, the lesion was solid and had a firm and fibrous texture, which make us consider the diagnosis of an ossifying fibroma or a FD (**Figure 6**).

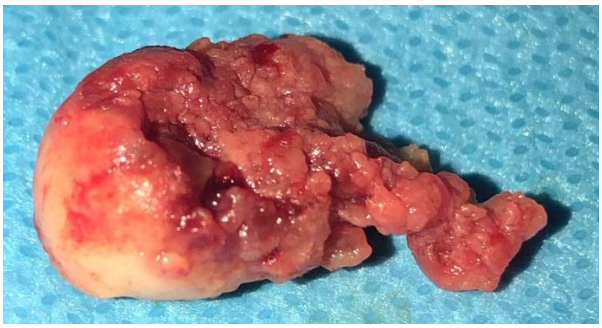

Figure 6: The specimen measuring 23mm along its major axis

The specimen was sent for histopathological examination, which confirmed the presence of a FD with an associated focal ABC.

At 3-month follow-up, the patient was asymptomatic (**Figure 7**). Another appointment for osteototmy has been scheduled.

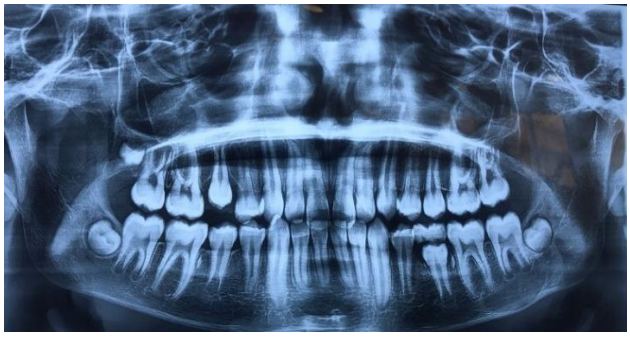

Figure 7: 3-month radiological follow-up

3. Discussion

FD accounts for about 2.5% of bone tumours and nearly 7.5% of benign bone conditions.[2,6,10] It is more frequent in young patients within the first three decades of life, and affects predominantly afro-descendent women.

The monostotic form accounts approximately 70-80% of cases and involves most frequently the rib, femur, jawbones, tibia, humerus (in decreasing order).[2] The polyostotic variant represents 20-30% of cases and affects the lower half of the skeleton.[6]

Craniofacial bones are involved in 30% of all cases of FD and seen in 10% of monostotic cases. Kaynak, with a higher prevalence in maxilla.[1,2,6]

The exact etiology is poorly understood, and various theories have been suggested. According to some researchers, local trauma and infection may contribute to the development of these conditions.[6] Other evidence believed that mutation in the guanine nucleotide-binding protein coding gene in early stages of life is responsible for the onset of the disease.[2,3,5,6]

Early-stage lesions are asymptomatic and are often discovered incidentally after radiographic evaluation. Swelling and asymmetry are the most common clinical sign of craniofacial FD.[5,6] Sometimes Teeth are displaced, rotated, maligned producing a severe malocclusion, and pain are observed.. We can say that it's a disease that significantly impairs the person functionally and aesthetically.

Other clinical features depend on the affected area, such as nasal obstruction, orbital dystopia with proptosis and diplopia with and without visual impairments.[6] In our case the patient's main complaint was swelling and facial asymmetry.

The radiographic presentation of FD is variable and depends on the stage of maturity of the lesion. Early-stage lesions appears as a radiolucent (cyst-like appearance) becoming more radiopaque as more bone is formed,[5,6] and presenting a ground glass or orange peel radiological pattern.[2,5,6]

Mandibular involvement lead to expansion of both lingual and buccal plates and sometimes bulging of the lower border.[3,6] And cystic lesions of considerable size lead to thinning and remodeling of the cortex, with a rare perforation of the cortex or periosteal reaction, as seen in our case.

ABC is extremely rare the head and neck area, accounting for 2% of all cases. The mandibule and maxilla are the most prevalent sites involved.[9,0]

It's more frequent within the first three decades of life, with a peak incidence between 10 to 20 years of age, and no gender predilection.[12] These finding are similar to those of our case.

The etiopathogenesis of this disease remains unclear. It can arise de-novo, or secondary, in association with an underling primary bone lesion such as the FD, the ossifying fibroma, the central giant cell granuloma, osteoblastoma, osteosarcoma, chondrosarcoma, chondromyxoid fibroma and others.[4,7,9]

Clinically, ABC presents with a cortical expansion, an erosion, a fibro-osseous matrix formation and sometimes a malignant-like tumor appearance.

This condition is classified into three types:

Conventional or vascular type (95%), caracterized by a rapid and expansive growth, a cortical perforation and a soft tissue invasion. Hitsologically, it presents with a multiple sinusoidal spaces filled with blood among fibrous stromal tissue, a multinucleated giant cells, an osteoid matrix and a hemosiderin deposits.

Solid type (5%): usually discovered by chance during a routine X-ray examination of due to mild asymptomatic swelling. Histological features comprise hemorrhagic areas, abundant fibroblastic and fibrohistiocytic elements with osteoclastic giant cells, zones of osteoblastic differentiation with osteoid and fibromyxoid calcified tissue.

Mixed type that combines both conventional and solid characteristics.[4]

Our patient presents with a mild painless, slowly progressing swelling, and the histological examination revealed hemorrhagic focus surrounded by giant cells, as well as fibroblastic areas, which is are consistent with the solid type. This case may be regarded as exceptionally rare due to its dual atypical characteristics.

The radiographic characteristics of ABC are variable and non-specific. ABC may be unilocular or multilocular, soap bubble or moth eaten radiolucency with defined or irregular border, and sometimes cortical perforation.[12]

The co-existence of FD and ABC is uncommon, with only 37 cases reported in the literature,[13,14] and only 6 involving the mandible (14). Rau et al. reported that FD with concurrent ABC is typically diagnosed in patients with an average age of 18.8 years, with a higher prevalence in men (60%) compared to women (40%).[14]

Several authors have tried to explore the association between the ABC and the FD, Some suggested that both of them my coexist at the same time, and others believed that one may develop from the other.[4] In our case, the histopathological analysis revealed the coexistence of FD with a focal area of ABC. This finding supports the second theory, that ABC can develop secondarily in association with a pre-existing FD.

The correct diagnosis for both of the lesions is based on clinical, radiographic and histopathological study. However, histopathological examination remains the most reliable method to confirm the diagnosis.

There is no consensus regarding the optimal treatment strategy. The treatment of FD and ABC is generally based on surgical management. It can vary from a curettage and peripheral osteotomy to a total resection and grafting to fill the bone void of the affected segment.[1,7,9,10,12]

The choice of the approach depends on various factors: age, rate of growth, severity of the symptoms, acute or progressive functional impairments such as alteration of sight, breathing, mastication or speech, cosmetic deformity and anatomic location location.[3,6]

Generally, FD becomes quiescent or slows down around puberty, so small lesions may only require biopsy to exclude other lesions, and routinary clinical observation.[6] In our case, we performed a conservative approach; excision of the lesion and curettage; and the patient is under observation.

The overall prognosis of ABC and FD is good.[15] The recurrence of primary ABC ranges from 10% to 50%; however, no cases of recurrence have been reported for secondary ABC.[13]

4. Conclusion

The coexistence of the FD and ABC is rare and poorly understood, making their diagnosis and treatment challenging for practitioners.

A thorough clinical, radiological, and histopathological assessment is essential for each case to differentiate these lesions and establish an appropriate and optimal treatment plan.

5. Patient Consent

Obtained.

6. Source of Funding

None.

7. Conflicts of Interest

The authors declare no conflicts of interest.

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