Unveiling a Rare Association: Aneurysmal Cyst and Fibrous Dysplasia of the Mandible - A Case Report

Case Report

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ABSTRACT

Introduction: Aneurysmal bone cyst (ABC) is a rare benign bone lesion, especially in the craniofacial region. Fibrous dysplasia (FD) is a benign skeletal disorder characterized by the replacement of normal bone with fibro-osseous tissue. The coexistence of ABC and FD in the mandible is extremely rare, with few cases reported in the literature.

Case Report: We report the case of a 25-year-old patient with a history of fibrous dysplasia of the right mandible who presented with a painful swelling in the parasymphyseal region. Imaging revealed a multilocular cystic lesion in the right mandible extending from the first premolar to the third molar. Surgical enucleation of the cyst and resection of the involved teeth were performed. Histopathological examination confirmed the diagnosis of fibrous dysplasia with secondary aneurysmal bone cyst. Postoperative recovery was uneventful, except for transient hypoesthesia of the inferior alveolar nerve, which resolved after 11 months.

Discussion: The association of ABC and FD is extremely rare, with fewer than 30 reported cases in the literature. This case highlights the importance of early diagnosis and appropriate surgical management to prevent complications and ensure optimal outcomes. The combined approach of enucleation and careful resection allowed for successful treatment, with histological confirmation of both conditions.

Conclusion: This case emphasizes the need for awareness of rare associations between benign bone pathologies such as ABC and FD. A multidisciplinary approach, including radiological and histological evaluations, is crucial for accurate diagnosis and appropriate treatment.

Key Words: bone cyst, fibrous dysplasia, mandible .

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INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign bone dystrophy, which occurs exceptionally in the jaws. It can be primary or secondary to a preexisting bone tumor. Fibrous dysplasia (FD) is a benign skeletal disorder, in which abnormal fibro-osseous connective tissue replaces medullary bone.

There are two forms of FD: a monostotic form, localized in only one bone and a polyostotic form.

In maxillary bones, FD occurs almost exclusively in its monostotic form. The association of ABC and FD is extremely rare, especially in craniofacial FD and existing literature comprises solely case reports or case series, collectively involving fewer than 30 cases.

CASE REPORT:

A 25-year-old patient with a known history of fibrous dysplasia of the right mandible presented to our department with a chief complaint of a painful swelling in the right parasymphyseal region of the mandible, which had been progressively evolving over the past month. Upon examination, an enlarged, firm and tender mandibular swelling was palpable in the right parasymphyseal region of the mandible.

The overlying skin appeared normal, with no signs of inflammation or ulceration. Intraoral examination revealed slight swelling of the mucosa in the right inferior premolar and molar area.

Sensitivity of inferior alveolar nerve was conserved. For a further evaluation, panoramic radiography and dentascan were requested. Radiographic examination revealed a well-defined multilocular cystic lesion in the right parasymphyseal region of the mandible, extending from the first premolar to the third molar and blowing out the external and internal cortical bones . [Fig 1 & 2]

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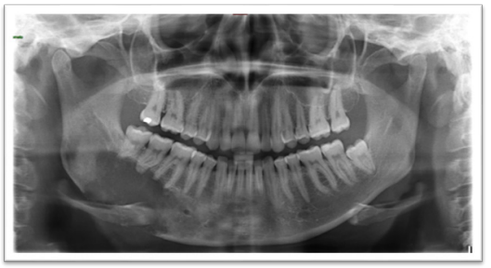

Figure 1 : Panoramic X-Ray showing a well-defined cystic lesion in the right parasymphyseal region of the mandible, extending from the first premolar to the third molar and blowing out the external and internal cortical bones.

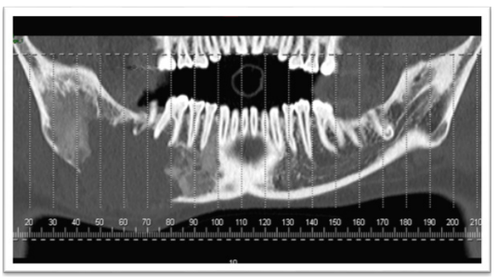

Figure 2 : Dentascan showing the same lesion and its contact with the roots of the right mandibular molars.

Under general anesthesia, we performed the enuclation of the clear fluid-filled cyst and apically resected the teeth. The inferior alveolar nerve was protected and pushed downwards. After enucleation, we filled the residual cavity with PRF. It was approximately 6 cm long and extended across the full width of the mandible, with the basilar rim blowing out both cortical bones. Histological examination confirmed the diagnosis of FD with secondary ABC. There were no signs of malignancy.

After surgery, the patient developed hypoesthesia of the inferior alveolar nerve, which gradually recovered after 11 months.

DISCUSSION

Aneurysmal bone cysts are rare, benign primary bone tumors that can be fast growing and locally destructive [1]. They are viewed as a secondary vascular phenomenon superimposed on an existing pathological process of the bone [2]. They account for 1-4% of all of all benign bone tumors [3]. Furthermore, ABC of the maxillae represent only 1.5% of non-odontogenic cysts [4].Aneurysmal bone cysts of the mandible are treated surgically [5], either by a conservative approach involving enucleation of the cyst and careful curettage of the cavity, or by a radical approach involving resection of the lesion with reconstruction. Fibrous dysplasia is a benign skeletal condition initially described by Lichtenstein, which involves the abnormal replacement of medullary bone with fibro cellular tissue due to altered fibroblast development [6, 7]. It accounts for 2.5% of bone tumors and 7% of benign bone tumors [8].

In the craniofacial region, FD is relatively uncommon, representing just 20% of all cases, and it almost exclusively presents in its monostotic form [9].

Fibrous dysplasia manifests as a gradually hardening bone swelling with normal skin and surrounding tissues. Alternatively, it may remain asymptomatic for an extended period and be incidentally discovered during an X-ray examination. Surgical treatment options for FD include three approaches [9].

Remodeling Surgery, used for extensive lesions where complete excision would result in excessive mutilation;

-Large excision recommended for small lesions with functional symptoms and for recurrences after previous remodeling surgery;

-Conservative management involving regular follow up, considered when there are no functional symptoms, in polyostotic forms, and during non-progressive phases in adolescents. This was the treatment option considered for our patient, who had mandibular FD prior to the appearance of the ABC.

The association of these two conditions is extremely rare. In 1975, Levy et al. published a study of 57 cases of ABC with other osseous lesions, where they did not encounter a single case of ABC with FD [10]. Another review published by Martinez and Sissons in 1988 found only one case of ABC in 42 patients with FD [11]. The preferred treatment for FD with ABC is surgical resection [12]. Therefore, we opted for surgery. The result was satisfactory for our patient.

CONCLUSION:

The association between two benign bone pathologies is possible, such as FD and ABC despite the rarity of its occurrence. Diagnosis must be based on clinical, radiological and anatomopathological arguments, in order to provide the patient with appropriate management.

CONFLICT OF INTEREST

the authors declare that there are no conflict of interest.

REFERENCES:

1- Zileli, M., Isik, H.S., Ogut, F.E. et al. (2013) 'Aneurysmal bone cysts of the spine', Eur Spine J, 22(3), pp. 593–601.

2- Kransdorf, M.J. and Sweet, D.E. (1995) 'Aneurysmal bone cyst: Concept, controversy, clinical presentation, and imaging', AJR Am J Roentegenol, 164, pp. 573-580.

3- Sokolo, R., Alseriani, F., Lagrange, A.C., Dumollard, J.M., Peoc'h, M., & Seguin, P. (2006) 'Kyste osseux anévrysmal du condyle mandibulaire', Rev Stomatol Chir Maxillofac, 107, pp. 109-114.

4- Perrotti, V., Rubini, C., Fioroni, M., & Piattelli, A. (2004) 'Solid aneurysmal bone cyst of the mandible', Int J Pediatr Otorhinolaryngol, 68, pp. 1339-1344.

5- Kiottavorncharoen, S., Joos, U., Brinkschmidt, C., & Werkmeister, R. (2003) 'Aneurysmal bone cyst of the mandible: a case report', Int J Oral Maxillofac Surg, 32, pp. 419-422.

6- Lichtenstein, L. (1965) Bone tumors. The CV Mosby.

7- Casselman, J.W., De Jonge, I., Neyt, L., De Clercq, C., & D'Hont, G. (1993) 'MRI in craniofacial fibrous dysplasia', Neuroradiology, 35(3), pp. 234-237.

8- Garau, V., Tartaro, G.P., Aquino, S., & Colella, G. (1997) 'Fibrous dysplasia of the maxillofacial bones: Clinical considerations', Minerva Stomatologica, 46(10), pp. 497-505.

9- Gangloff, P., Polo, A., Moizan, H., Froment, N., & Gerard, E. (2004) 'Tuméfaction maxillaire révélatrice d'une dysplasie fibreuse. A propos d'un cas', Med Buccale Chir Buccale, 10(2), pp. 77-81.

10- Levy, W.M., Miller, A.S., Bonakdarpour, A., & Aegerter, E. (1975) 'Aneurysmal bone cyst secondary to other osseous lesions: Report of 57 cases', Am J Clin Pathol, 63(1), pp. 1-8.

11- Martinez, V. and Sissons, H.A. (1988) 'Aneurysmal bone cyst: A review of 123 cases including primary lesions and those secondary to other bone pathology', Cancer, 61(11), pp. 2291-2304.

12- Swed, S., Kremesh, M.I., Alshareef, L., et al. (2021) 'Secondary aneurysmal bone cyst with benign fibro-osseous lesions: Case report', Ann Med Surg (Lond), 72, p. 103024.