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Ameloblastic Fibro-Odontoma of the Mandible: Uncommon Case Report

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Case Report

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Abstract: Ameloblastic fibro-odontoma (AFO) is a tumor, which has the histologic features of ameloblastic fibroma (AF) in conjunction with the presence of dentin and enamel. It is less common than AF, accounting for 0.3–1.7% of jaw tumours, signifying its rarity. There is no gender or anatomic site predilection. We report a case of AFO in a 40-year-old female patient involving the left body of the mandible. After a promising presurgical evaluation, a conservative surgical approach, including enucleation of the lesion and curettage of the surrounding bone was done. To ovoid fracture of the mandible, splint fixation with circummandibular wires was used. Clinical and radiographic control, two years after surgery, showed perfect healing of the lesion and no sign of recurrence.
Keywords: Ameloblastic fibro-odontoma, odontoma, mixed odontogenic tumors, benign tumors

INTRODUCTION

The ameloblastic fibro-odontoma (AFO) is a rare benign mixed odontogenic tumor with the general features of ameloblastic fibroma but that also contains enamel and dentine [1].

It is defined by the World Health Organization (WHO) as a neoplasm composed of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that resembles dental papilla, with varying degrees of inductive change and dental hard tissue formation [2].

AFO is normally found in young patients with no significant gender predilection [1]. It occurs with equal frequency in the maxilla and mandible [3]. Clinically, AFO presents as a painless swelling of the affected area [1] Radiography shows a well-defined, radiolucent area containing various amounts of radiopaque material of irregular size and form [3]. The aim of this paper is to report an uncommon case of a large mandibular AFO in a female patient and reviews the relevant clinicopathological features of this neoplasm.

CASE REPORT

A 40-year-old female patient was referred to oral surgery department with a chief complaint of painless swelling of the left anterior region of the madibula present for two months. The clinical examination revealed a firm, asymptomatic swelling in the left mandible (Fig 1). There was no history of local trauma or infection. The initial panoramic radiograph revealed a well-defined, radiolucent lesion, which contained several radiopaque bodies of varying sizes and shapes extending from the right central incisor (41) to the left second premolar (35) while weakening the basal bone of the mandible (Fig 2a). The cone beam computed tomography (CBCT) sections confirmed the presence of a large mixed lesion involving the buccal and lingual cortical plates of the left mandible with local destruction of the buccal bone (Fig 2 b, c and d). Considering the clinical and radiological picture the possible differential diagnosis was AFO, immature complex odontoma, calcifying epithelial odontogenic tumour, and calcifying odontogenic cyst. Under local anesthesia, the mass was removed by enucleation followed by bone curettage(Fig 3).

To ovoid fracture of the mandible, splint fixation with circummandibular wires was used. Macroscopic examination of the specimen revealed a hard tissue mass with a soft tissue attachment. The histopathologic examination of the surgical specimen showed cords and small islands of odontogenic epithelium in a loose primitive-appearing connective tissue that resembled the dental papilla. In addition, it

showed conglomerate mass of enamel and dentine arranged in a disorganized pattern and in close relationship to the ameloblastic epithelium(Fig 4).

There was no evidence of malignancy and the final diagnosis was of AFO. Two years after surgery, there were no signs of recurrence and the clinical and radiological appearances of the bone and surrounding soft tissue were normal (Fig 5).

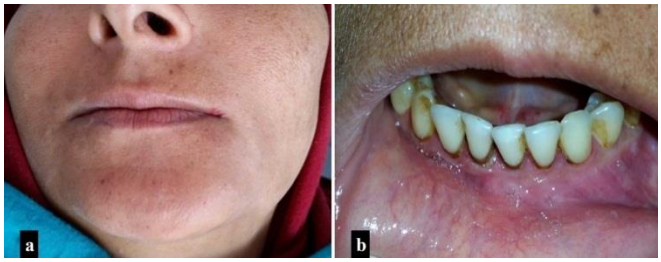

Fig-1: Clinical examination showing slight swelling of the mandibular anterior region

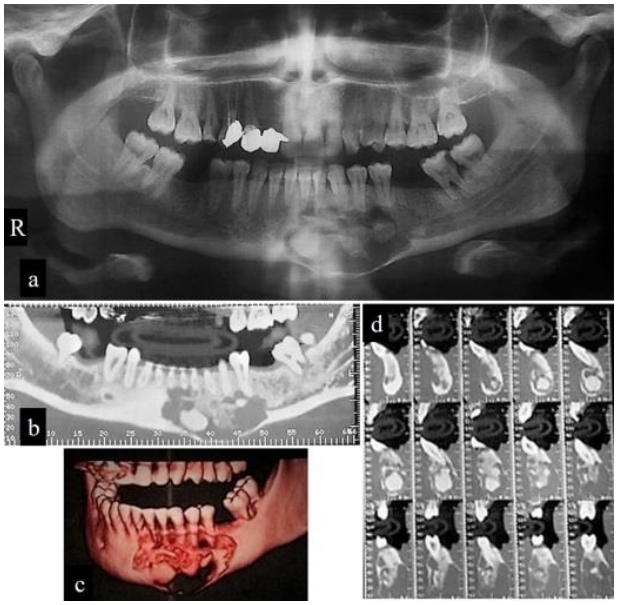

Fig-2: Initial panoramic radiograph showing the lesion in the left anterior region of the mandible (a). Cone beam computed tomography: Panorex (b), three-dimension reconstruction (c) and Coronal-oblique reconstructions showed mixed mandibular lesion (d).

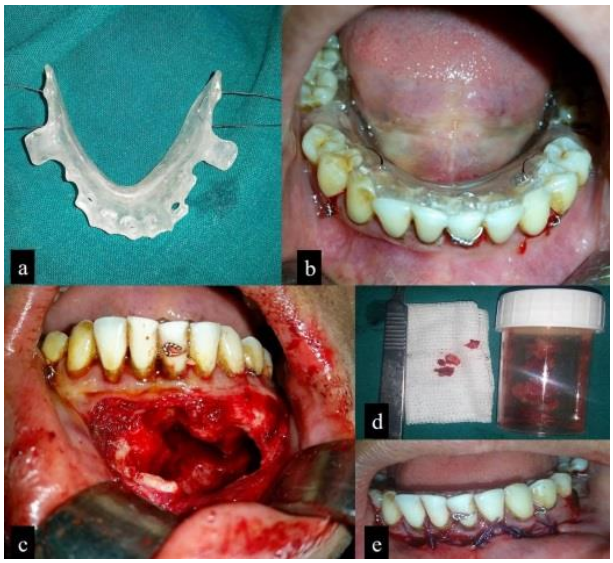

Fig-3: Splint fixation with circummandibular wires (a and b), Surgical enucleation of the lesion (c), specimen (d) and Sutures (e).

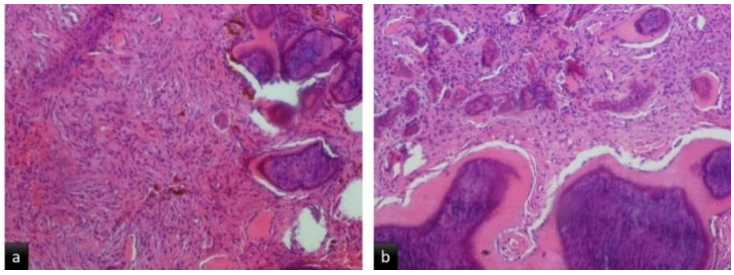

Fig-4: Chords of odontogenic epithelium in a loose primitive appearing connective tissue that resemble dental papilla (left) and a tooth-like structure (right) (a). Decalcified section showing areas of enamel and dentine in close relationship with the ameloblastic epithelium (b)

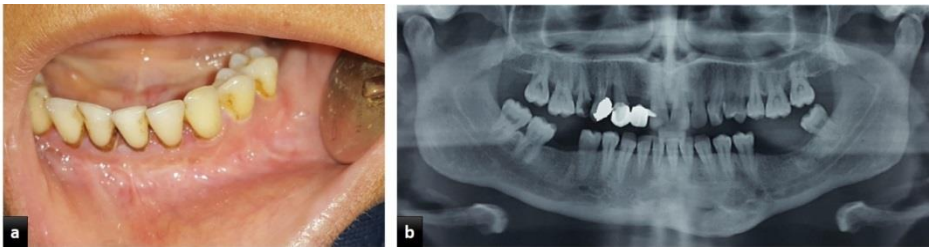

Fig-5: Clinical and radiographic examination two years after surgery showing reossification and no recurrence of the lesion

DISCUSSION

AFO is relatively uncommon, with a prevalence of 1–3% among odontogenic tumors [3, 4]. There is no gender predilection, with the lesion being equally found in the mandible and maxilla [5]. Generally it is seen in the first and second decades of life, which might also be a characteristic of the lesion. However, AFO may also occur at advanced ages as reported in our case [1]. There has been a lot of discussion in the literature regarding its proper classification. One point of discussion is the discrimination between neoplasm and hamartoma [3, 6]. Recently, according to the 4th Edition of the World Health Organization (WHO) Classification of Head and Neck Tumors 2017, the decision was made to group AFO and ameloblastic fibro-dentinoma (AFD) under odontomas as developing odontomas. It was concluded that there was little evidence to justify classifying AFD and AFO as independent entities [7].

Common clinical and radiological signs and symptoms of AFO are asymptomatic swelling, which occasionally can produce facial disfigurement, as well as a well-circumscribed unilocular or multilocular radiolucency with varying levels of radiopacity depending on the extent of mineralization [1, 2, 4]. Nevertheless, final diagnosis is made according to microscopic evaluation demonstrating islands of odontogenic epithelium embedded in cell-rich ectomesenchyme similar to dental papilla [1, 3, 8]. Dentin and enamel matrix are also seen [5]. Differential diagnosis of AFO should include lesions with mixed radiographic patterns, such as calcifying epithelial odontogenic tumour, calcifying odontogenic cyst, immature complex odontoma and adenomatoid odontogenic tumor [5].

The treatment of AFO is enucleation or curettage without removing the adjacent teeth since it is considered a non-aggressive lesion [3, 6, 9]. Sporadic recurrences of AFO have been attributed to the inadequate surgical enucleation of the lesion or if tumor remnants are retained in the resected margins, especially in cases with large tumors [1, 10].

Malignant transformation is rare, nevertheless has also been reported. Hence, long-term follow-up is recommended [5]. When there is recurrence accompanied by changes in the histological pattern towards a more unorganized fibrous stroma with displacement of the epithelial component, more extensive treatment procedures are indicated [3].

CONCLUSION

We have reported a case of a large AFO that was presented as asymptomatic swelling of the left anterior region of mandible. The lesion was treated conservatly as a bengin tumor by enucleation but they need long-term follow-up due to the possibility of recurrence and malignant transformation.

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