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Research Article

AMELOBLASTIC FIBRO-ODONTOMA OF THE MAXILLA: CASE REPORT OF AN UNCOMMON ODONTOGENIC TUMOR

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A R T I C L E I N F O

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A B S T R A C T

Ameloblastic fibro-odontoma (AFO) is an extremely rare benign hybrid odontogenic tumor (OT), occurring in the early stages of life. AFO normally presents as a painless swelling in the posterior of mandible or maxilla with mixed radiological appearance within well-defined borders. The treatment of this entity includes enucleation of the tumor and long-term follow up.

This article reports the first case of an ameloblastic fibro-odontoma to be reported in a moroccan military medical institution and reviews the literature regarding the clinical, radiological and histopathological features of this neoplasm.

INTRODUCTION

AFO is a rare benign mixed odentogenic tumor (OT). It was first described by Hooker in 1967, as a separate lesion from ameloblastic odontoma (AO) [1, 2]. It is a slow-growing usually asymptomatic tumour that occurs in young patients with no significant sex predilection [3, 4].

Clinically it presents as a swelling of the mandible or maxilla, with destruction of cortical bone. Radiologically, the AFO appears as a uni or multilocular radiolucency with a well defined borders. Histopathologically, AFO is composed of odontogenic epithelium immersed in embryogenic connective tissue that mimics primitive dental pulp [1, 3].

It is the purpose of this paper to report the first case of AFO in a moroccan military medical institution.

Case Presentation

A 32-year-old patient was referred to the oral and maxillofacial surgery department of our institution complaining of diffuse tumefaction of the right mandibular area that is evolving since 6 months. There was no history of systemic disease and trauma.

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The tumefaction expanded slowly, without other symptoms. Oral examination revealed an asymptomatic swelling on the right side of the mandible without signs of inflammation. The buccal and lingual osseous corticals expansion was seen. No cervical lymphadenopathy was noted.

Panoramic radiograph shows unilocular radiolucent lesion with well-defined borders involving in the right body of the mandible, extending anteriorly to the right first premolar, with a very thin cortex and resorption of first molar roots (Figure 1).

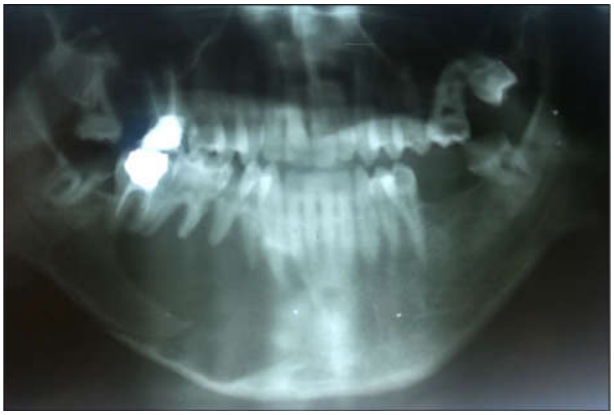

Figure 1 Panoramic radiograph shows unilocular radiolucent lesion with well-defined borders involving in the right body of the mandible, extending anteriorly to the right first premolar

Computed tomography (CT) and 3D-CT confirmed the presence of a large radiolucent mass in posterior body of mandibular ramus (Figure 2 & 3).

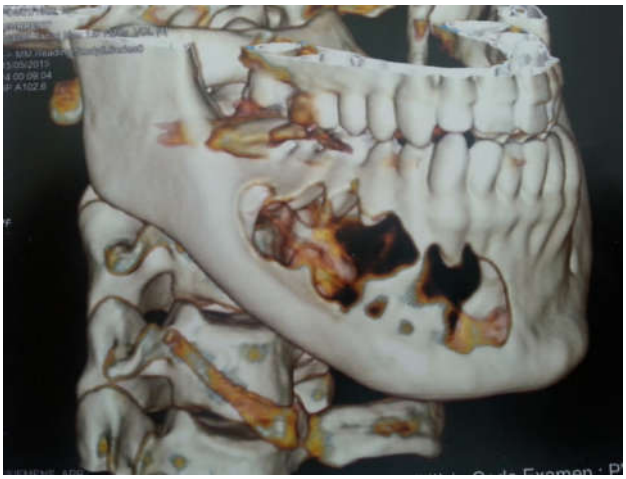

Figure 2 Three-dimensional computed tomography showing the lesion of the right mandible.

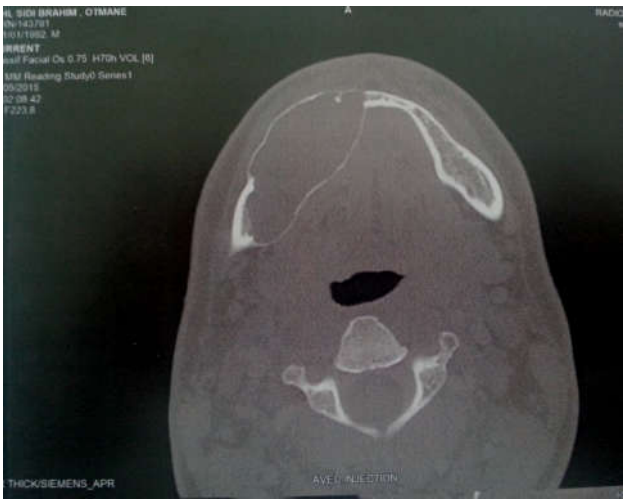

Figure 3 Computed tomography showing large radiolucent mass on right mandible.

Excision of the mass was performed through extra-oral approach, and the specimen was submitted for histopathological analysis. The tumor measured 6x3,8 cm with a smooth surface.

Macroscopic section of the tumor showed both cystic and solid component, gray white colored viscous liquid was seen in the cystic cavities.

Microscopic examination showed anastomotic cords and small islands of odontogenic epithelium in a loose primitive appearing connective tissue that resembled the dental papilla (Figure 4). The epithelial islands show peripheral columnar cells surrounding a looser collection of spindle cells while the stroma is cellular, composed of stellate and spindle shaped cells (Figure 4). Throughout the lesion, the products of odontogenesis were readily identified: mature tubular dentin and enamel matrix (figure 5). There was no evidence of malignancy, including no nuclear pleomorphism, and the tumour was diagnosed as an AFO.

Eight months after surgery, there were no signs of recurrence and the clinical and radiological appearances of the bone and surrounding soft tissue were normal.

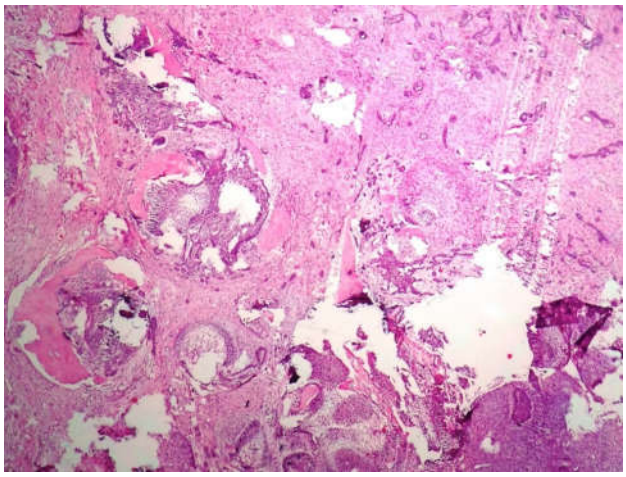

Figure 4 Photomicrograph of ameloblastic fibro-odontoma showing strands, cords and nests of odontogenic epithelium supported by richly cellular connective tissue (HE, x50)

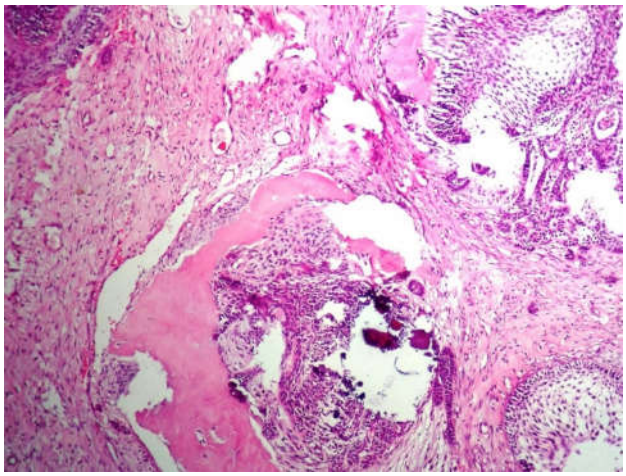

Figure 5 The soft tissue component of the tumor intermixed with the calcified products of odontogenesis tissue (HE, x100)

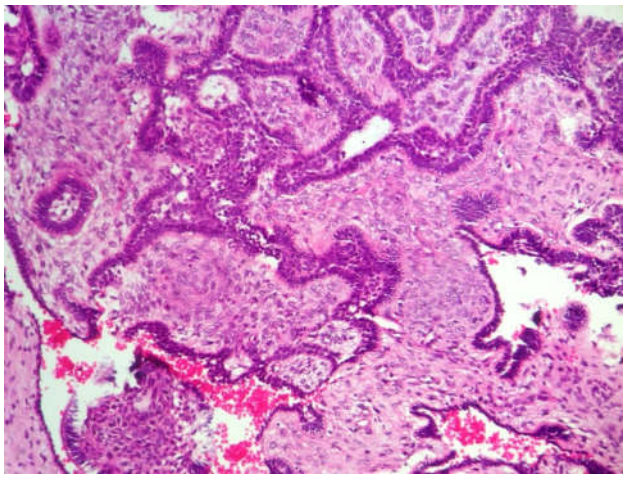

Figure 6 The islands of ameloblastic epithelium in an ectomesenchymal stroma.

DISCUSSION

AFO is a rare benign OT defined by World Health Organization (WHO) as a mixed neoplasm composed of proliferting odontogenic epithelium embedded in cellular ectomesenchyme tissue with varying degrees of inductive change and dental hard tissue formation [1, 3].

Although it was described as a distinct entity in the mid-1960s,

some controversy persists as to whether the AFO represents a distinct lesion or is merely a stage in the development of an odontoma. A significant proportion of AFOs seem to grow to a much larger size compared with the odontoma, suggesting that at least some of these tumors represent a separate and distinct lesion from the odontoma [2].

There are different concepts about the nature of AFO in the literature. The term "epithelial odontogenic tumour with odontogenic mesenchyme" is becoming more widely accepted these days and avoids potential controversy over the nature of the neoplasia. The term "ameloblastic fibro-odontoma" represents a histologic combination of ameloblastic fibroma (AF) and complex odontoma [3,4].

On the basis of inductive principle, the development of mineralized components in OT is a sequel of epithelial and mesenchymal interactions in which the ameloblastic epithelium stimulates differentiation of odontoblasts from mesenchyme to deposit dentin. In turn, this induces the formation of enamel matrix. AFOs shows a complete interaction between epithelial and mesenchymal components and consequently both enamel and dentin are formed [4,5].

AFO is relatively uncommon and has been reported with a prevalence range of (0–3.4%) within OTs among different regions. Generally, it is seen in the first and second decades of life, which might also be a characteristic of the lesion. Hooker reported the mean age of patients as 11.5 years (range 6 months to 39 years). However, AFO may also occur at advanced ages [2,6,7].

The common site is the posterior mandible, with a mandible–maxilla ratio of 2:4 [2,4,7]. The most common clinical presentations of AFO are asymptomatic swelling and failure of tooth eruption without altering their vitality [4].

Radiologically, AFO shows a circumscribed multilocular or unilocular radiolucent area and a center comprising a radiopaque material of irregular size and form with various amounts, depending on the maturity of the odontogenic hard-tissue structures. Some of the lesions are relatively small when first detected, measuring 1 to 2 cm in diameter, whereas others may be exceedingly large, involving a considerable portion of the body of the mandible or maxilla [7,8].

Histopathologically, AFO is generally surrounded by a fibrous capsule and is composed by strand, cords, and islands of odontogenic epithelium immersed in moderately cellular embryoginic connective tissue, that mimies primitive dental pulp.

The epithelium is characterized by peripheral palisading of columnar cells that surround loose spindled epithelium, resembling stellate reticulum.The epithelial component shares many features of ameloblastoma however, the stroma is strikingly different. The stroma is a cellular ectomesenchyme made up of spindle-shaped cells that resemble the dental papill Formation of osteodentin and enamel is also observed microscopically) and represents the feature that separates the AFO from ameloblastic fibroma. As with several other odontogenic lesions, benign melanocytic colonization of the epithelial component has been reported in the AFO [4,8-11].

With respect to histogenesis, some controversy surrounds the relationship between ameloblastic fibroma, ameloblastic fibrodentinoma (AFD), AFO and odontoma. Some consider them as separate entities. Others regard them as chronological stages in a continuum beginning from ameloblastic fibroma at one extreme and odontoma at the other extreme with AFO as well as AFD in an intermediate stage [5,12,13,14].

The AFO shares similarities with the AF and the AFD but may de distinguished from these two entites by the presence of dentine and enamel on histological examination [10].

However, the concept that these lesions represent a continuum of differentiation is not widely accepted, and others feel that they are separate pathologic entities [13,14,15].

The recommended treatment of AFP is conservative surgery with enucleation or curettage as it does not appear to locally invade the bone AFO are reported to recur only rarely. When the lesion includes an unerupted tooth, the tooth should be removed with the mass. Unlike ameloblastic fibroma, AFO is not generally associated with a malignant transformation to ameloblastic fibrosarcoma. This difference in malignant potential further supports the separation of these two tumors [4,7,15]

Our case is distinguished by the young age of the patient, the size of the tumor and the absence of recurrence after surgical resection.

CONCLUSION

AFO is a rare benign neoplasm with very low incidence of malignant change. Its diagnosis remains a challenge for oral pathologists. There are many lesions which mimic ameloblastic fibro-odontoma clinically and histopathologically. These conditions must be ruled out before making a definitive diagnosis of ameloblastic fibro-odontoma.

Competing Interests

The authors declare that there is no conflict of interest regarding the publication of this paper.

List of abbreviations

AFO: Ameloblastic fibro-odontoma
AFD: Ameloblastic Fibro-dentinoma
OT: Odontogenic Tumor
AF: Ameloblastic Fibroma

Ethics approval and consent to participate

Not applicable

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