Ameloblastic fibro-odontoma of the maxilla: a case report

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The ameloblastic fibro-odontoma is a rare benign odontogenic lesion defined as a tumor with the general features of ameloblastic fibroma but that also contains enamel and dentin. In this article the authors describe a case of a young male patient with ameloblastic fibro-odontoma of the maxilla and the management of such condition.

Keywords:
ameloblastic fibro-odontoma, ameloblastic, fibromaodontogenic tumor

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Introduction

The ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic lesion defined as a tumor with the general features of ameloblastic fibroma but that also contains enamel and dentin [1]. According the recent WHO classification of odontogenic tumors published in 2005, AFO belongs to the group of lesions with odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation [1].

AFO is normally found in young patients, with no significant sex predilection. The incidence of AFO is between 1 and 3% in odontogenic tumors [2,3].

Clinically, it presents as a painless swelling of the affected area, usually the posterior portion of the maxilla or mandible. Radiographs show a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form [4–7]. Conservative surgical excision is the treatment of choice, and the lesion does not tend to recur.

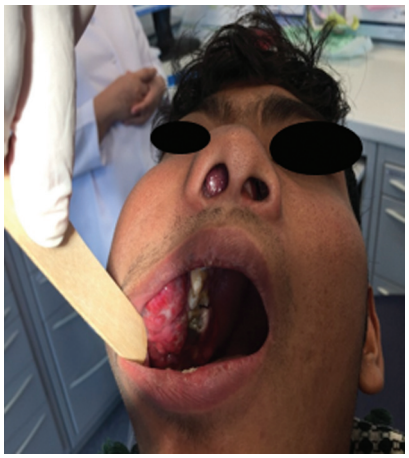
Report of the case

A 24-year-old man who was otherwise fit and healthy presented to the accident and emergency department with facial swelling on the right side for 2 months. The patient was referred to the department of oral and maxillofacial surgery for further evaluation. The patient was initially complaining of mild pain, facial deformity, change in voice, and mild visual disturbance (Figs 1 and 2).

Histopathological finding

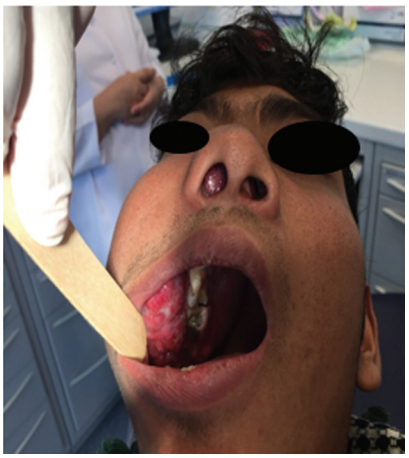
Incisional biopsy showed surface squamous epithelium and entrapped cords of odontogenic epithelium within

Figure 1



Facial deformity and intraoral lesion with impingement on the left nostril.

Figure 2



Facial deformity and intraoral lesion with impingement on the left nostril.

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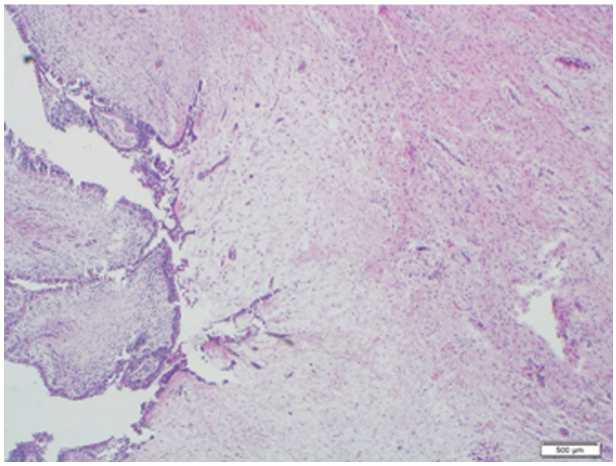
slightly myxoid stroma. The stroma exhibits mild-to-moderate cellularity and is composed of bland spindle to angulated cells. The odontogenic epithelium exhibits peripheral columnar cell palisade and central reticulum-like cells. All of the above features are suggestive of AFO. Figures 3 and 4 show microscopic features described above.

Radiological finding

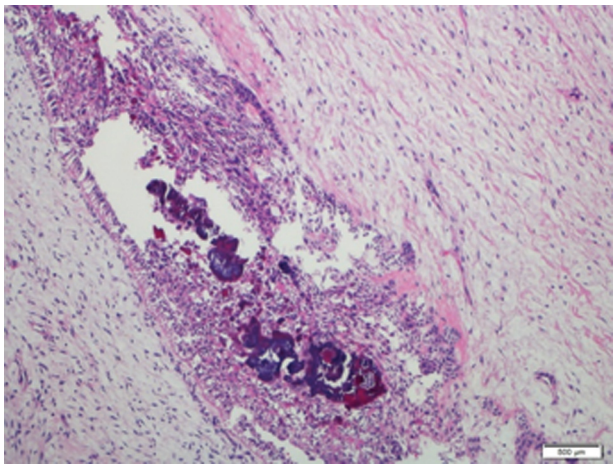
A computed tomography scan was requested, which showed a massive destructive lesion occupying the entire maxilla and zygoma and extending from the alveolar bone on the right side to the orbital floor and from the zygoma to the right nostril suggestive of ameloblastoma (Figs 5 and 6). MRI was taken to assess the soft tissue extension.

Figure 3



Entrapped cords of odontogenic epithelium within slightly myxoid stroma. The stroma exhibits mild-to-moderate cellularity and is composed of bland spindle to angulated cells.

Figure 4



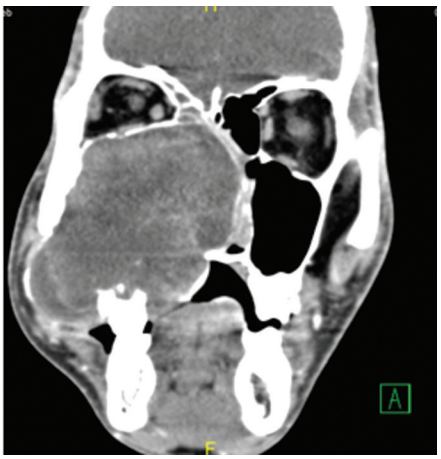
The odontogenic epithelium exhibits peripheral columnar cell palisade and central reticulum-like cells. Areas of calcification are noted in association with the epithelium.

Treatment plan

Measurements were taken for obturator to be used perioperatively and postoperatively, and the patient was admitted for local excision and reconstruction with obturator.

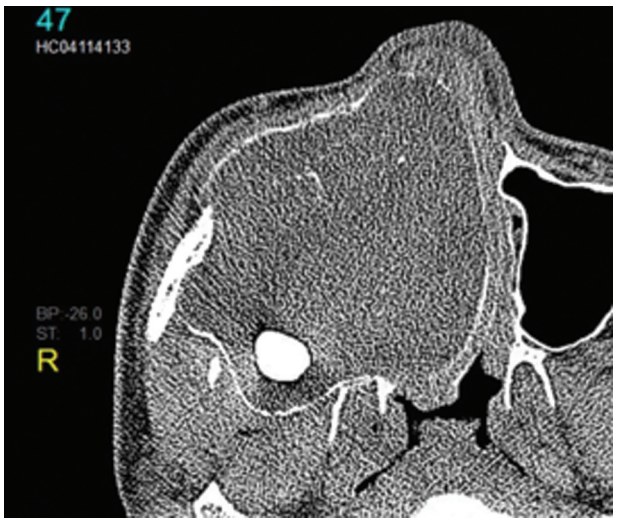
Weber Ferguson incision with Dieffenbach's modification was used to approach the massive tumor. After exposure, the lesion was found to be well capsulated and no bone removal or further excision beyond the capsule was deemed necessary. The entire tumor was removed without rupture. Nasal mucosa seen bulging in the images was intact. Obturator constructed preoperatively was placed and a nasojejunal tube was inserted. Patient recovery was unremarkable. Several postoperative outpatient visits showed the resolution of all symptoms and with final prosthesis the patient is currently back to

Figure 5



Coronal view of the lesion.

Figure 6



The tooth within the lesion.

his lifestyle without complication or signs of recurrence (Figs 7–9).

Discussion

Hooker [8] first differentiated the AFO from the ameloblastic odontoma. If AFO, ameloblastic fibroma, and odontoma are simply stages in a continuum, clinical data on each of lesions should support this – for example, the ameloblastic fibroma should occur in younger patients, the odontoma in somewhat older patients, and AFO in an intermediate age group.

Slootweg [5] has investigated this issue and concluded that AFO represents a separate specific neoplastic entity, and AFO should not be considered as hamartomatous as there are cases of AFO showing true neoplastic behavior and the existence of malignant variants [9].

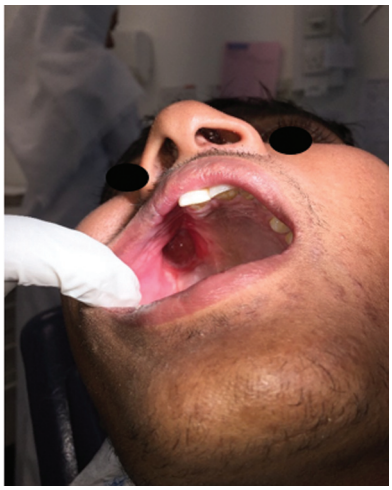
Now most investigators agree that AFO is a separate entity. The revised WHO classification of odontogenic tumors defines AFO as 'a lesion similar to ameloblastic fibroma, but also showing inductive changes that lead to the formation of both dentin and enamel' [10].

Some investigators believe that this entity represents an immature form of odontoma and therefore should be regarded as a hamartoma rather than a true neoplastic process. In some cases, however, the tumor can undergo progressive growth, causing bone destruction and significant deformity, as seen in the present case. Such destructive lesions seem to be true neoplasms. Most of the other lesions may simply represent a stage in odontoma development. However, differentiation cannot be made solely on a histopathologic basis.

A review by Philipsen *et al.* [6] in 1997 reported 86 cases of AFO in the literature. Of those 86 cases, only one case had been reported with an age at diagnosis of over 20 years. All other cases presented in children with an age range of 1–22 years and an average age at presentation of 9 years. In the case presented here, the age of the patient was 24 years, which is not coinciding with findings of the previous study by Philipsen and colleagues.

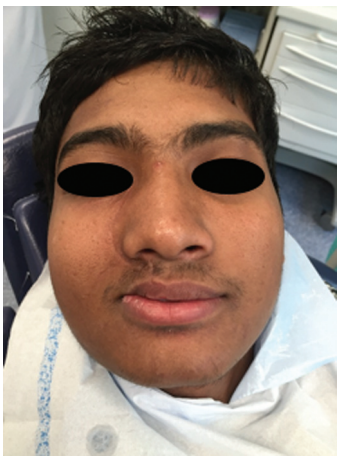
Philipsen *et al.* [6] noted a negligible male to female ratio of 1.4 : 1. The majority of AFOs were found in the posterior mandible, with the mandible affected more than the maxilla by a ratio of 2 : 4.

Figure 7



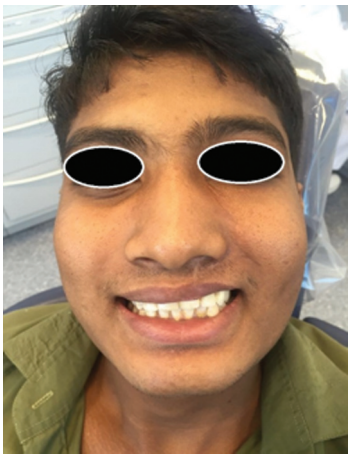
The oral cavity 2 weeks postoperatively.

Figure 8



Facial form and prosthesis in function.

Figure 9



Facial form and prosthesis in function.

Radiography usually shows a well-defined radiolucent area containing various amounts of radiopaque material

of irregular size and form. The ratio of radiopaque-to-radiolucent areas differs from one lesion to another; sometimes, the mineralized element in the tumor predominates and the lesion may resemble an odontoma [11]. Some of the lesions are relatively small when first detected, measuring 1–2 cm in diameter, whereas others may be exceedingly large, involving a considerable portion of the body of the mandible or maxilla [12].

In the case presented here, computed tomography showed a massive destructive lesion occupying the entire maxilla, zygoma, and the alveolar bone on the right side and bending the orbital floor, and bulging into the right nostril. Within the lesion there was a radiopaque round mass, which was consistent with a molar crown.

When AFO presents with the typical age, location, and radiographic pattern, the diagnosis is usually obvious.

Histologically, the tissue masses of an AFO show characteristic structure of an immature complex odontome consisting of irregularly arranged enamel, dentinoid, cementum, and pulp-like ectomesenchymal tissue [13,14]. Strands and island of odontogenic epithelium with peripheral columnar cells resembling ameloblast and central stellate reticulum-like cells [14–16], embedded in typical rich ectomesenchyme, were also observed.

This lesion is generally treated by conservative enucleation and curettage. The tumor is well circumscribed and does not invade the surrounding bone; therefore, it tends to separate easily from the surrounding bony cavity. The prognosis for this lesion is excellent and recurrence is rare. The few reported recurrences may be attributed to inadequate removal of the entire lesion [17–19]. Therefore, it is essential to provide adequate access to ensure complete removal of the lesion, especially around the apices of erupted dentition [20]. Long-term follow-up is recommended.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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