Med Buccale Chir Buccale 2017;23:99–102
DOI: 10.1051/mbcb/2016055

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Up-to Date Review and Case Report

Report of a rare case of ameloblastic fibro-odontoma of the mandible with >400 odontomas and a related literature review

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(Received: 11 August 2015, accepted: 30 October 2016)

Keywords: ameloblastic fibro-odontoma / mandible / enucleation

Abstract – Introduction: Ameloblastic fibro-odontoma (AFO) is a benign odontogenic tumor of mixed origin. It is often asymptomatic and can be diagnosed by a delayed eruption of a permanent tooth. AFO is often associated with one or more impacted teeth. **Observation:** The patient in the reported case was a 20-year-old girl, who consulted us because of pain associated with functional impairment in the left mandibular region for approximately 4 years. Intraoral examination demonstrated bud-like lesion at the location of the missing left lower molars. The computed tomography scan showed intralesional calcifications and dental densities. Enucleation was performed under general anesthesia. Excised tissues included two teeth, with their pericoronal bags, and hundreds of microelements isolated or embedded in membranes. Macroscopic examination showed >400 odontomas, and 28 membranous portions containing other odontomas, apart from permanent teeth. Postoperative recovery was satisfactory. **Conclusion:** It is an extensive odontogenic tumor often associated with an impacted tooth.

Introduction

Ameloblastic fibro-odontoma (AFO) is a benign odontogenic tumor of mixed origin [1] with a dual component – odontogenic epithelium and odontogenic ectomesenchyme close to the dental papilla. It represents 0.1–3.4% odontogenic tumors and develops most commonly between age 8 and 12 years [2]. It is often associated with one or more impacted teeth. This work reports a case of AFO in the mandible containing >400 odontomas.

Observation

The patient in the reported case was a 20-year-old woman who consulted us for pain associated with left-side mandibular functional discomfort developing over a period of 4 years. She mentioned a history of dental avulsions and repeated infection. She also gave a history of digestive problems with recurrent constipation and hemorrhoids from an early age. Palpation revealed left mandibular lymphadenopathy.

Extraoral examination showed that the face was asymmetrical, with a small folded swelling on the left cheek.

Intraoral examination demonstrated a bud-like hemorrhagic lesion at the location of absent teeth 35, 36, 37, and 38, retaining the print of the opposing teeth that had erupted (Fig. 1). Masticatory function was disturbed. The outer cortex showed expansion. An orthopantomogram revealed dense, punctate microelements in the mandibular body and left branch, along with two impacted teeth (Fig. 2).

The scanner showed a left mandibular oblong formation of 6 cm × 2 cm with the presence of intralesional calcifications and bone densities. The cortex was expanded without any cortical discontinuities (Fig. 3).

Enucleation was performed under general anesthesia. A trapezoid incision was made with a supracrestal cut with an anterior release in relation to the canine and a posterior release toward the mandibular branch. After detachment, enucleation involved two impacted teeth with their follicular

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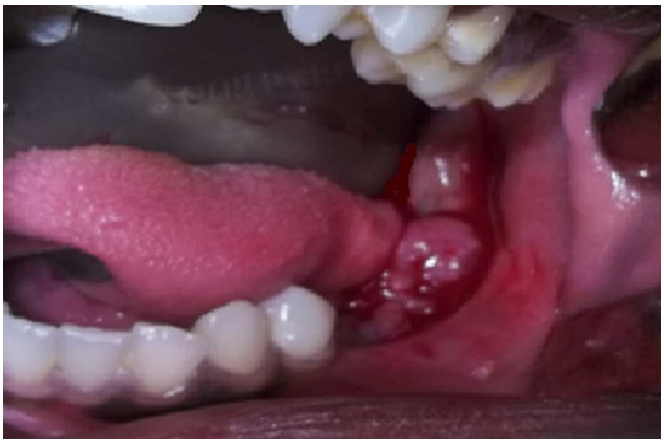

Fig. 1. Hemorrhagic budding lesion.

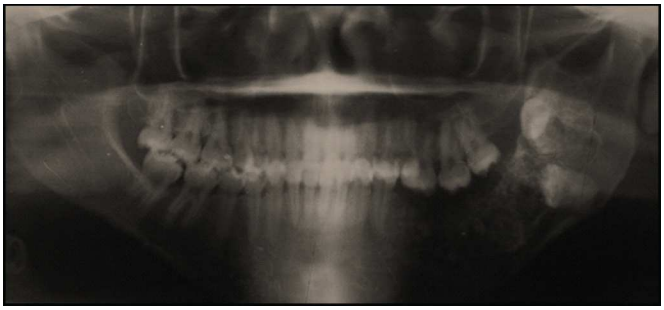

Fig. 2. Multiple osteo-condensation with two impacted teeth.

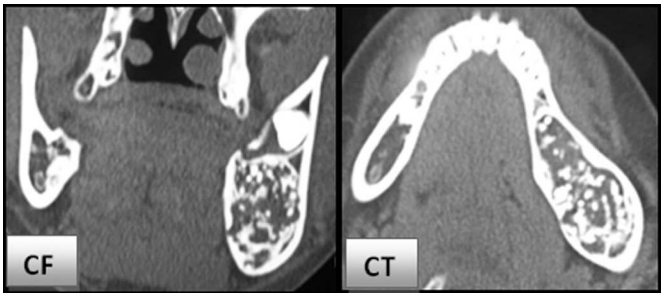

Fig. 3. Intralesional calcifications with expanded cortical, frontal (CF) and transverse (CT) sections.

sac, hundreds of microelements either isolated or embedded in membranes. Bony lacunae had limited our access to and made enucleation of some odontomas difficult (Fig. 4).

Thus, apart from the impacted teeth 47 and 48, surgery involved membranes containing the odontomas and the other multiple isolated odontomas (Fig. 5). Macroscopic examination showed approximately 400 isolated odontomas, and 28 membranous portions with other odontomas (Fig. 6).

At microscopic examination, the fragments showed a multitissue proliferation consisting of a cubic–cylindrical odontogenic epithelium forming several small acini and spans of abundant fibrocollagenic deposition with hard elements (enamel and dentin; Fig. 7).

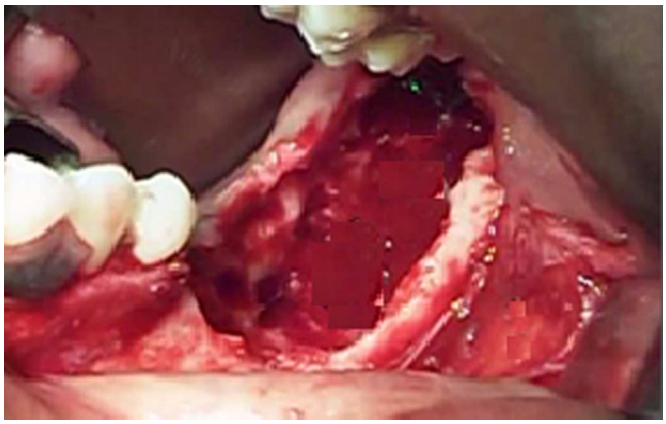

Fig. 4. Tumor cavity after enucleation, with medial cortical bony lacunae.

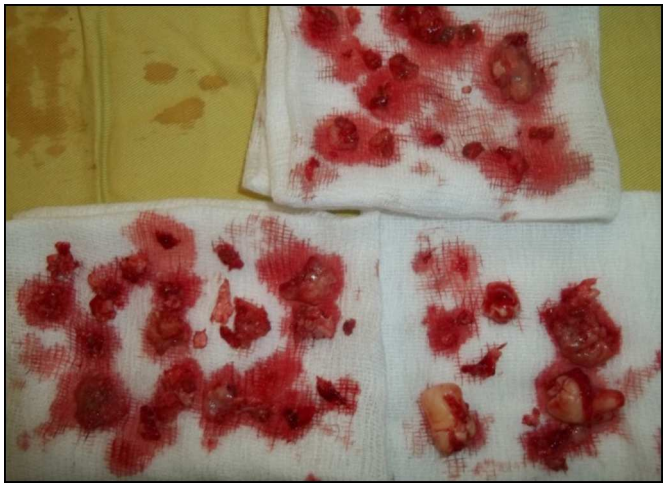

Fig. 5. Specimen with membranes, multiple odontomas, and teeth 37/38.

Postoperative recovery was satisfactory with only mild labiomental hypoesthesia, which faded 3 months afterward. During follow-up evaluations, the mucosa was found to be closed and the orthopantomogram did not show any abnormalities (Figs. 8 and 9).

Comments

AFO is classified as odontogenic epithelium with odontogenic ectomesenchymal tumors, with or without the formation of hard tissue [1]. This tumor is diagnosed in 71% cases between ages 5 and 14 years. The average age of diagnosis is 9.6 years, with a range of 8 months to 26 years [3]. Age is therefore an important component of differential diagnosis [4–6]. Buchner [3] found a male:female ratio of 1.85 (65% men). This tumor is often located in the posterior mandibular region (54% [3] and 58% [7]).

Late consultation often leads to an aggressive expansion to the basilar edge and mandibular branch [7]. In the case reported herein, a delay in consultation is the result of poor

Fig. 6. Over 400 odontomas identified with membranes containing other odontomas and teeth.

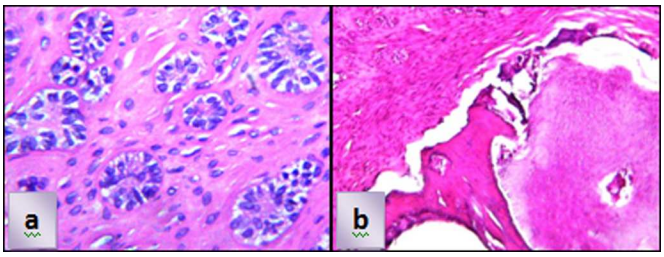

Fig. 7. Histopathology: odontogenic epithelium with fibrocollagenic proliferation (a) dental hard tissue and (b) (HE ×100).

socioeconomic conditions, ignorance, the painless character of the lesion, and the lack of experienced specialists in oral surgery.

AOF is often asymptomatic and diagnosed as a result of a delayed eruption of one or several permanent teeth (83–91.6% with swelling of the affected area [2,3,5,7,8]. In 29% cases, the discovery of this tumor is incidental on X-rays taken after a delayed eruption of one or several teeth [3], as in the case reported here. Radiologically, it has the appearance of a large isolated bone cyst, generally centered by a radio-opaque lesion whose rim is punctuated by a pattern or cluster more or less confined with very dense opacities, and dental color [4,7,9]. The single-cyst (unilocular) form represents 90.3% and the radio-opaque form is associated with a bright radio-opaque lesion in 94.8% cases [3].

With an average age of tumor occurrence between 8 and 12 years and three components (fibroma, ameloblastoma, and odontomes), AOF is different from other odontogenic epithelium with odontogenic ectomesenchyme tumors with or without hard-tissue formation. These tumors are as follows: ameloblastic fibroma (average age of 14.8 years) ameloblastic fibro-dentinoma, calcified odontogenic epithelium tumor, adenomatoid odontogenic tumor, odonto-ameloblastoma, and odontomas (around the age of 40 years) [3,4,7,10].

At the microscopic level, the odontogenic epithelium will proliferate on a follicular or plexiform template within a stroma of mature connective tissue, similar to the findings in classic ameloblastoma. Hard dental structures similar to compound or complex odontomas are observed in these epithelial proliferations [11–13].

The recommended treatment consists of conservative enucleation with curettage [4,5,14]. Impacted teeth must be extracted so as to limit the risk of recurrence [4]. In the case

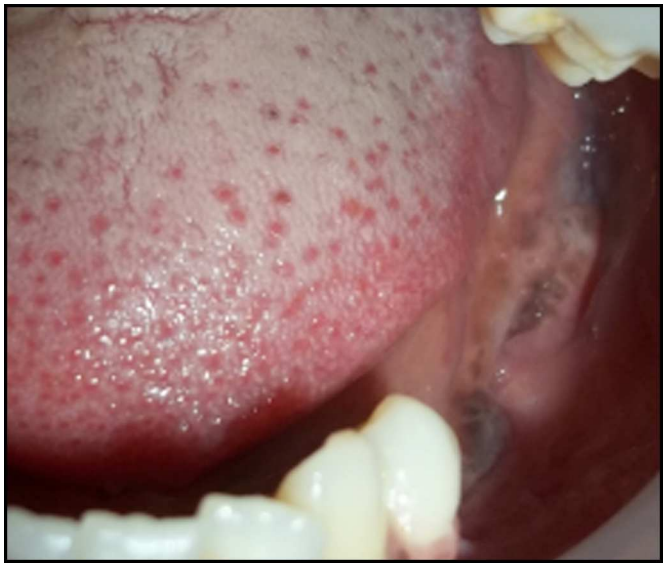

Fig. 8. Follow-up intraoral photograph at 3 months after surgery.

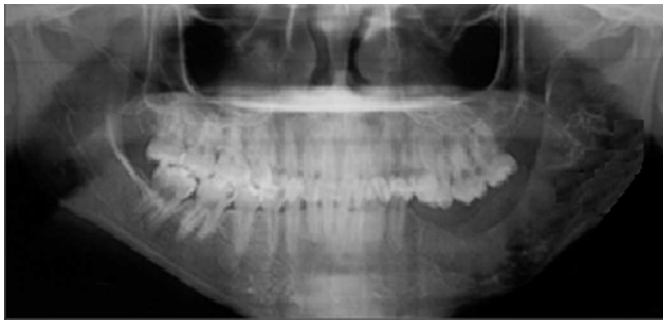

Fig. 9. Follow-up radiograph at 3 months after surgery.

reported above, the bony lacunae rendered access to and enucleation of the approximately 400 embedded odontomas difficult. Such a high number of odontomas (400 isolated elements) reported in this clinical observation is very rarely found in the literature.

Recurrences are very rare after full enucleation [4]. However, four cases of recurrence have been reported [15,16].

Conflicts of interests: The authors declare that they have no conflicts of interest in relation to this article.

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