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Case Report

Ameloblastic fibro-odontoma: Case report and immunohistochemical profile

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ABSTRACT

Ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic tumor of epithelial and ectomesenchymal origin associated with formation of mineralized products of odontogenesis. This manuscript reports an AFO affecting the posterior mandible of a 10-year-old boy with detailed clinical, radiographic and histopathological evaluation. The incisional biopsy was performed which revealed strands and islands of odontogenic epithelium composed of columnar peripheral cells surrounding central region where the cells were loosely arranged, resembling the stellate reticulum of enamel organ. The mesenchymal portion consisted of numerous ovoid and stellate cells in a loose matrix, similar to dental papilla in development. At this point, the diagnosis was ameloblastic fibroma (AF). The patient underwent complete surgical excision of the lesion associated with application of Carnoy's solution and maintenance of the first molar. The histopathological analysis established the final diagnosis of AFO, since the same histopathological characteristics of AF were observed plus enamel and dentin matrix material on close relationship with epithelial structures. Immunohistochemical panel (AE1/AE3, CK14, CK19, Vimentin, β-catenin, S-100, Ki67) was performed to illustrate AFO features. Treatment of odontogenic tumors is based on the biological and clinical behavior. In general, prognosis is excellent, with rare recurrence reports and even rarer reports of malignant transformation. No recurrence or signs of other tumors have been observed in the patient for 1 year after tumor resection.

1. Introduction

Ameloblastic fibro-odontoma (AFO) is a rare odontogenic tumor of mixed epithelial/mesenchymal origin, comprising 1–3% of all odontogenic tumors [1]. Tumors in the head and neck rarely affect children; however, odontogenic tumors are frequent in childhood and adolescence. AFOs usually arise in the first two decades of life, with no significant gender or ethnic predilection [2,3].

AFO classically presents as a painless swelling in the posterior portion of maxilla or mandible and frequently is associated with unerupted teeth [3]. Therefore, AFO is often noticed in routine radiographs taken because of teeth eruption failure [4]. Diagnosis of AFO is based on histological evidence of a biphasic tumor composed of odontogenic epithelium proliferation within a highly cellular mesenchymal tissue with a primitive appearance, but also containing tooth-like structures as enamel and dentin in different degrees of maturation throughout the tumor [5,6].

The literature describes AFO as a non-aggressive tumor generally treated by conservative surgical enucleation, with occasional reports of expansive and locally destructive progression [6,7]. Immunohistochemical data concerning AFO in the current literature are very limited and is a reflex because of the rarity of this lesion. The present report adds a new case of AFO with detailed immunohistochemical information and illustrates the clinical, radiological and histopathological features.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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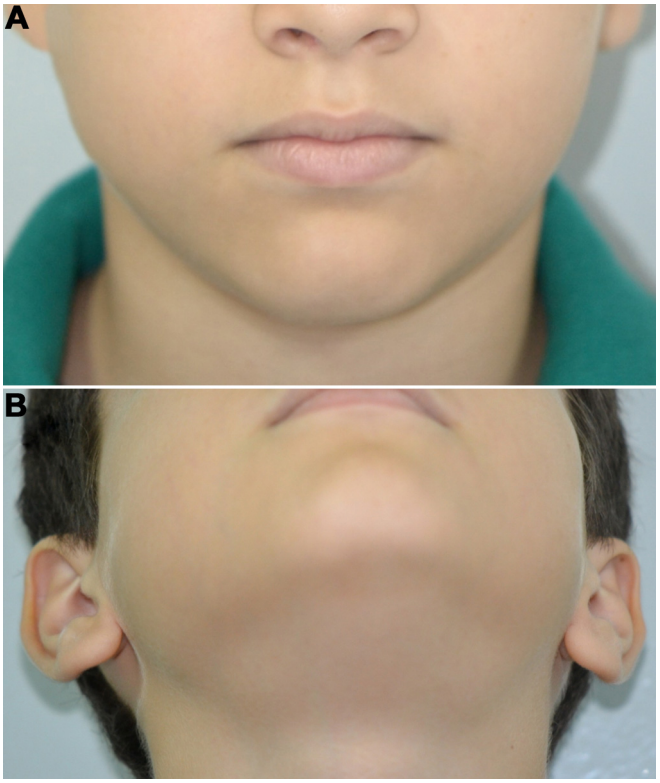

Fig. 1. Mild facial asymmetry on the right side of the jaw (A and B).

2. Case report

A 10-year-old boy presented to a private dental clinic for evaluation of a slight swelling in the face with duration of 4 weeks. On physical examination, a mild facial asymmetry was evident on the right side of the face (Fig. 1). Intraoral examination revealed a marked enlargement on the right posterior mandible measuring approximately 4.0 cm in greatest diameter, normochromic and resulting in expansion of cortical bone (Fig. 2). Medical history was non-contributory. Panoramic radiograph showed a well-circumscribed, radiolucent, unilocular lesion containing radiopaque material in its interior and affecting the right mandibular body, angle and ramus (Fig. 3). Tooth germs of second and third right mandibular molars were absent and the lesion exhibited relationship with the tooth root of the first mandibular molar. The diagnostic hypothesis of the surgeon was

Fig. 2. Intraoral view. A swelling was observed in the mandible (right).

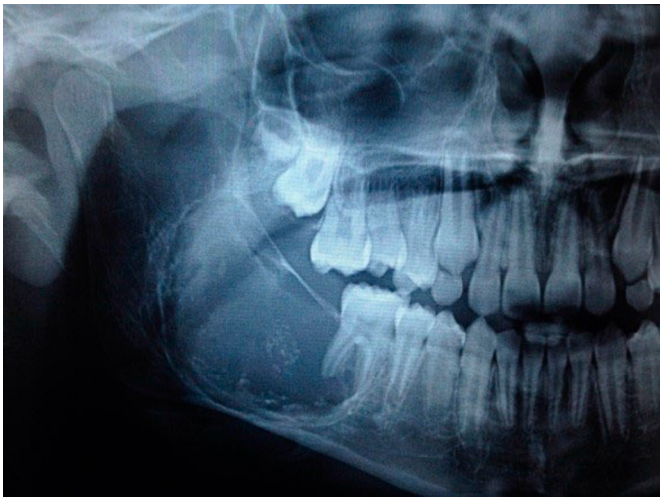

Fig. 3. Details of panoramic radiograph showing radiolucent unilocular lesion in right mandible with radiopaque areas inside.

ameloblastic fibroma (AF). Aspiration of the lesion was negative for the presence of liquid. Next, an incisional biopsy was performed and the specimen was referred to our department for histopathologic analysis, which revealed strands and islands of odontogenic epithelium (Fig. 4) composed of columnar peripheral cells surrounding central region where the cells were loosely arranged, resembling the stellate reticulum of enamel organ. The mesenchymal portion consisted of numerous ovoid and stellate cells in a loose matrix, similar to dental papilla in development. At this point, the diagnosis was AF. The patient underwent complete surgical excision of the lesion associated with application of Carnoy's solution and maintenance of the first molar. The specimen was fixed in 10% formalin solution and the histopathological analysis established the final diagnosis of AFO, since the same histopathological characteristics of AF was observed plus enamel and dentin matrix material on close relationship with epithelial structures (Fig. 5). A complementary immunohistochemical panel was performed on3-μm thick deparaffinized tissue sections. Table 1 exhibits the specifications of the primary antibodies (anti-AE1/AE3, anti-CK14, anti-CK19, anti-Vimentin, anti-β-catenin, anti-Ki67 and anti-S-100). After antigen retrieval, endogenous peroxidase was blocked with a 1:1 solution of methanol and 3% hydrogen peroxide. Sections were incubated with the primary antibodies and treated with the labeled streptavidin biotin complex (LSAB + System-HRP; Dako, Carpinteria, CA). The reaction was developed with diaminobenzidine as chromogen, counterstained with Mayer's hematoxylin and mounted in

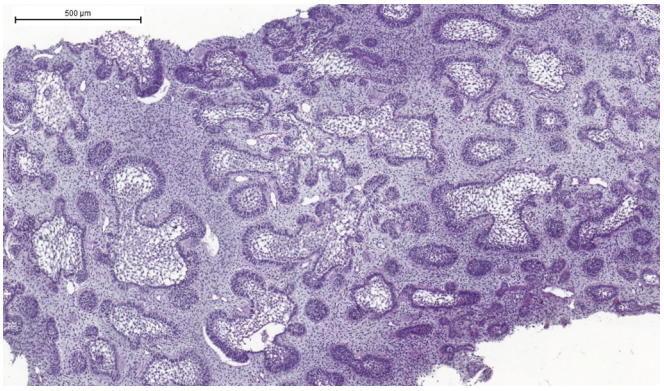

Fig. 4. Photomicrograph of the biopsied specimen revealing cords and islands of odontogenic epithelium embedded in loose primitive connective tissue highly cellularized and consistent with an ameloblastic fibroma (H&E).

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