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Up-to Date Review And Case Report

A rare case of maxillary ameloblastic fibro-odontoma and a review of the literature

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Abstract – Introduction: Ameloblastic fibro-odontoma (AFO) is a benign complex odontogenic tumor in the mandible in children and adolescents. **Observation:** A 9-year-old boy was referred to the dental department for the delayed eruption of tooth 26. He was asymptomatic, and on clinical examination, we found that tooth 26 was absent with tumefaction instead of the tooth. Cone beam computed tomography revealed an opaque mass in the root of 26. The management was surgical, and anatomopathological examination facilitated the diagnosis of AFO. **Commentary:** The upper maxillary localization of AFO is rare and is not often a practitioner's first thought. Surgical excision of the tumor is essential based on the histological diagnosis. The patient must be followed up after 3 months and regularly until the eruption of the affected tooth. **Conclusion:** AFO is one of the differential diagnoses to be considered when a young patient presents with firm swelling with delayed eruption of the opposing tooth, especially since it has good prognosis and is easy to manage.

Introduction

The ameloblastic fibro-odontoma (AFO) belongs to the family of mixed odontogenic tumors, representing 2% of these tumors [1]. It is mainly found in young patients, with a male predominance, and it is localized preferentially around the mandibular molars. It is generally asymptomatic, and the diagnosis can be made following a delay in dental eruption.

Observation

A 9-year-old patient was referred by his dentist following the failed eruption of tooth 26 (Fig. 1). He had no childhood trauma history or any other specific history. The patient had no symptomatic complaints. On clinical examination, he presented an asymptomatic swelling on palpation opposite the missing tooth with a healthy mucosa. The alveolar bone was bulging on the vestibular face without mucosal intrusion. Orthopantomogram (Fig. 2) revealed a radiopaque multinodular lesion in the second quadrant. It was superimposed on the root of 26, which was difficult to isolate. The contours were blurred and poorly defined. In the first quadrant, tooth 16 was in place without issue on the arch, and the intramucosal root of 17 was impacted.

A cone beam computed tomography (Fig. 3) was performed to assess the anatomical relationships of the tumor with its neighboring structures, which revealed a well-defined radiolucent lesion with X-ray elements and dental inclusions. The planned management was surgical and performed under general anesthesia. After orotracheal intubation and infiltration of the vestibule of tooth 26 with adrenaline xylocaine, a crestal incision was made. The detachment of a mucoperiosteal flap revealed a tumor with a thick white wall. The maxillary cortical bone was lysed at the level of the lesion. Total excision was performed in a single attempt, carefully separating the tumor from its cavity (Fig. 4). The latter was curetted and rinsed to remove the entire lesion, taking care not to damage the root of tooth 26. The follicular sac of tooth 26 was visible at the base of the cavity. The lesion was sent for pathological analysis. Finally, the periosteal mucosal flap was sutured to obtain a hermetic closure.

The patient was seen again with his parents at the 3-week follow-up consultation. The mucosa was healed, and they were given the results. Histological analysis (Figs. 5 and 6) found a loose myxoid connective tissue comprising epithelial elements of odontogenic type, arranged in the form of spans and lobules. It also included mineralized zones. This aspect was suggestive of an AFO, and the diagnosis was confirmed by the reference laboratory in Paris.

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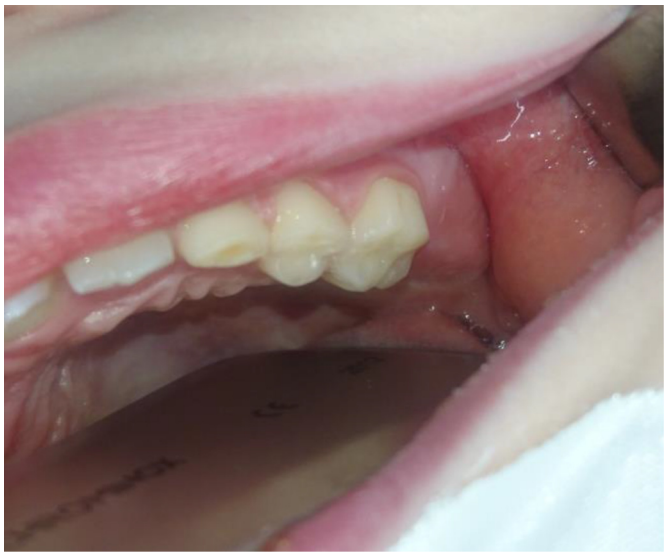

Fig. 1. Clinical view: Slight swelling of the vestibule at tooth 26.

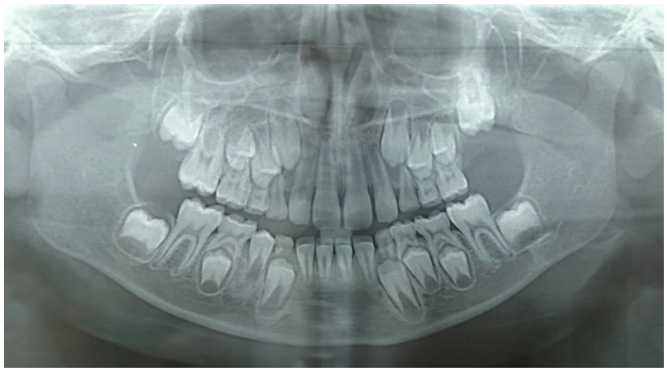

Fig. 2. Orthopantomogram: Tooth 26 is impacted with a mix of radiolucent and radiopaque lesions above.

Commentary

AFO is a rare tumor that can affect up to 3.4% of the population. It is mainly diagnosed in people under the age of 20 [2], with a slight predominance in men. This benign tumor initially develops asymptomatically. The mass leads to a delay in dental eruption, which pushes back the missing permanent tooth to the apex of its final position. Depending on the tumor growth, there may be edema next to the tumor, sometimes accompanied by pain. The main clinical sign being delay in tooth eruption [2], diagnostic explorations are quickly implemented, which may explain why the diagnosis is made quickly, before the age of 20. Radiological examinations help locate the tumor. In the decreasing order of frequency, AFO is often found in the posterior regions of the mandible [3], followed by the anterior region of the maxilla, including the incisors and canines [4]. In the present case, the location was the posterior region of the maxilla, which was less common.

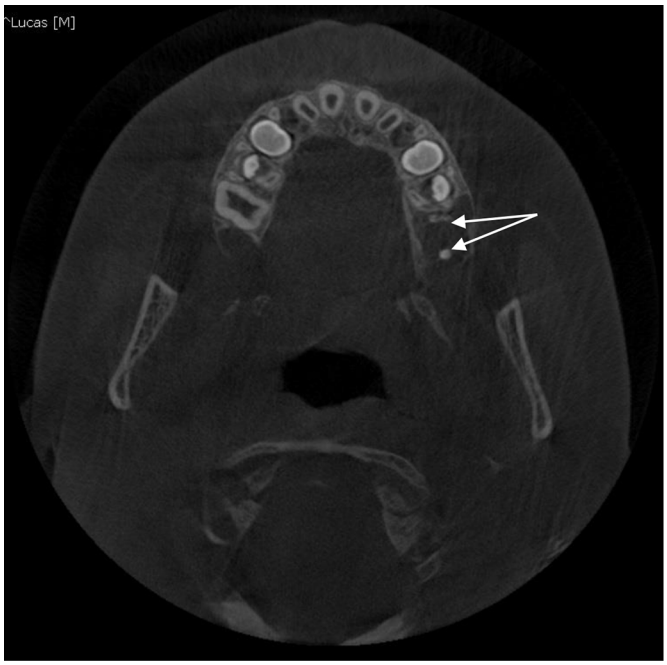

Fig. 3. Axial section of the cone beam: dental inclusion (white arrows) at the site of tooth 26.

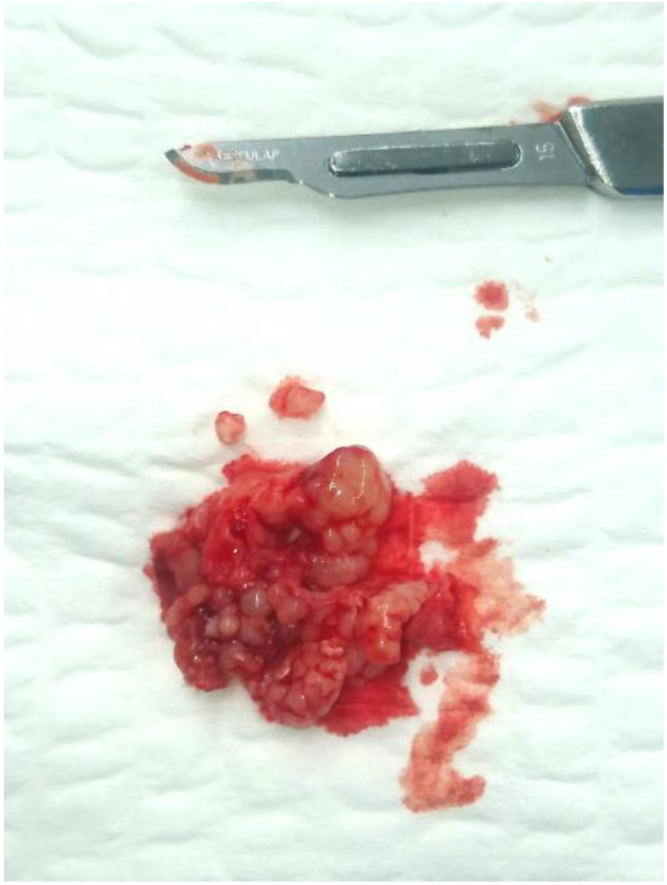

Fig. 4. Operatory piece: Lesion found inside the cavity.

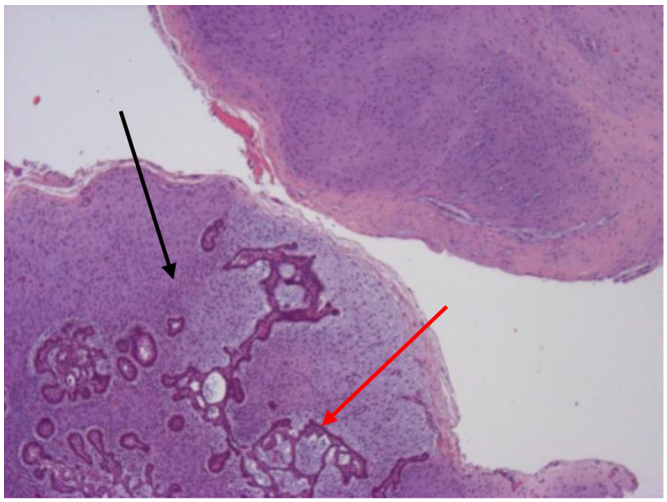

Fig. 5. Histological section. G × 50: Ameloblastic fibro-odontoma: Mesenchymal tissue (black arrow) with epithelial column (red arrow).

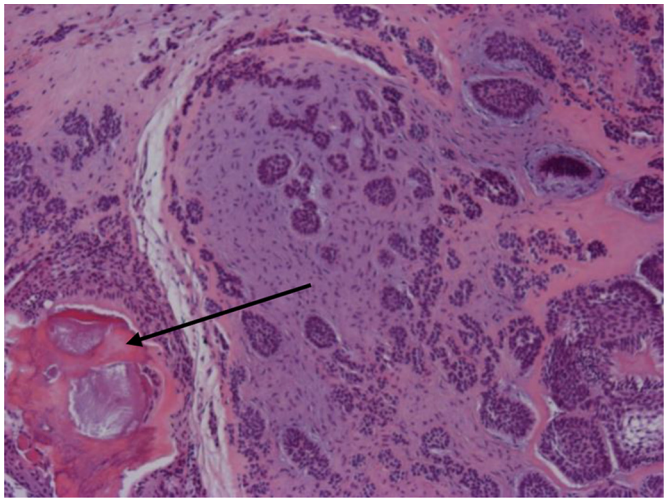

Fig. 6. Histological section. G × 100 : Ameloblastic fibro-odontoma. G × 100. Orange mineralized area (black arrow).

The use of cone beam computed tomography makes it possible to obtain good resolution imagery. It indicates the relationships of the lesion with neighboring anatomical structures, its volume in space, and the nature of the structures that compose it according to their density [5].

Imaging suggests several differential diagnoses: complex odontoma, compound odontoma, ameloblastic fibro-dentinoma, ameloblastic fibroma. The clinical signs are often the same for all these tumors. Only anatomopathological examination can produce an accurate diagnosis. AFO consists of epithelial spans on a mesenchymal background with mineralized elements. AFO is similar to ameloblastic fibro-dentinoma, but it contains amelar and dentinal materials, whereas ameloblastic fibro-dentinoma contains only dentinal materials. On the radiological level, AFO has more opaque and denser elements within the lesion than ameloblastic fibro-dentinoma, suggesting the presence of enamel [6]. Ameloblastic fibro-dentinoma radiologically differs from AFO with its multilocular and [7] not unilocular lesions. Ameloblastic fibro-dentinoma is made up of clusters of epithelial cells within a mesenchymal tissue simulating that of the dental papilla but does not contain mineralized elements. Odontomas are more common [3] and are categorized as follows: complex odontoma and compound odontoma. Complex odontoma shows an image of a radiopaque block on radiography, and odontoma is composed of a mass of dental sketches [3]. Histologically, odontomas contain all the dental elements (enamel, cement, dentin, and connective tissue pulp), unlike AFO. These elements are represented at a more advanced stage than AFO, with complete oral development in compound odontoma but with malformed elements [3].

Total excision of the lesion should be performed to avoid a recurrence or even a malignant transformation. Some authors [8,9] advise to remove the root affected by AFO. In the present case, the dissection and removal of the part allowed a monobloc excision. The follicular sac and the root of tooth 26 were visible during the procedure and were undamaged, so they were left in place. Howell and Burkes described two cases in which an AFO developed into ameloblastic fibrosarcoma [10], suggesting that the risk increases with relapses. It should be suspected in the presence of nuclear atypia and a loss of the epithelial component [11]. The diagnosis of AFO must also be made as early as possible because an overly large lesion can lead to more complicated surgery [12].

From a classification point of view, AFO was put in its own subclass in the family of complex mixed odontogenic tumors in 2005, according to the World Health Organization. It was said to be mixed because it contains mesenchymal and epithelial elements at the histological level. It was said to be “complex,” unlike “compound” tumors because the dental elements included in the mass are immature, disorganized, and do not yet resemble the final dental structures. Conversely, the elements of compound odontomas, more frequent than complex odontomas [4], consist of several small dental elements morphologically similar to a tooth.

Now, the new classification of 2017 [13], published by the World Health Organization has removed the names of AFO and ameloblastic fibro-dentinoma. It established that these two forms are stages in the development of odontomas before the production of mature dental tissue. They are therefore grouped in the family of odontomas.

Conclusion

During a delay in dental maxillary eruption in a young patient, a radiological assessment, if necessary supplemented by 3-dimensional imaging is necessary to explore the cause of this delay. In case of a hyperdense and well-defined multiloculated mass, complete surgical excision is required to study its histology. Despite a predominance of occurrence in the mandible, AFO should not be overlooked among the

etiologies of maxillary tumors. The ameloblastic fibro-odontoma is therefore a benign tumor whose follow-up is necessary after excision, until the eruption of the permanent tooth and the absence of recurrence. At present, this term is still used despite the updated classification of head and neck tumors.

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