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Ameloblastic fibro-odontoma associated with paresthesia of the chin and lower lip in a 12-year-old girl

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Abstract

Ameloblastic fibro-odontoma is a rare, benign, and slowly growing neoplasm of the jaw composed of proliferating odontogenic epithelium in ectomesenchymal tissue along with dental hard tissue formation. Herein, we describe a case of an ameloblastic fibro-odontoma in 12-year-old female with paresthesia of the chin and lower lip. Panoramic radiography showed a radio-opacity in the right posterior mandible near the mandibular canal and associated with the right mandibular third molar. Histologically, the lesion contained epithelial and mesenchymal odontogenic components in close proximity to odontoma-like elements. Enucleation and curettage of the affected site in the mandible resulted in resolution of the paresthesia postoperatively.

Keywords

Ameloblastic fibro-odontoma, ameloblastic fibroma, odontoma, odontogenic tumor, mixed tumor

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Introduction

Ameloblastic fibro-odontoma (AFO) is an uncommon, well-encapsulated, slow-growing, and expansile benign odontogenic lesion.[1] It is usually diagnosed in the first two decades of life, with over 90% of cases occurring before the age of 20 years with a slight male predilection.[2,3] Patients with AFO usually present with a painless swelling of the posterior region of the mandible or maxilla, causing delayed eruption, displacement, or loosening of the involved teeth. The lesion can cause bony expansion and, subsequently, facial asymmetry. By radiologic imaging, the lesion usually appears as a well-demarcated, predominantly radiolucent area, containing variable amounts of radio-opaque material of irregular form and size with a density similar to that of hard dental tissue, or as a unilocular mixed lesion associated with the crown of impacted tooth.[3] On occasion, the radio-opacity can predominate, resembling a complex odontoma.

In 2005, the World Health Organization (WHO) defined AFO as a neoplasm composed of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that is similar to the dental papilla.[4] AFO is an odontogenic tumor with histopathologic features of ameloblastic fibroma (AF) in conjunction with the presence of some combination of dentin, enamel, and cementum that displays varying degrees of mineralization.[5] Treatment is usually surgical enucleation along with removal of any involved teeth. Recurrence is uncommon after adequate surgical removal.[6]

Case report

A 12-year-old female presented to our institution with a radio-opaque right posterior mandibular lesion associated with the right mandibular third molar found by her general dentist on routine dental radiographs. The patient reported an intermittent tingling sensation within the right side of her

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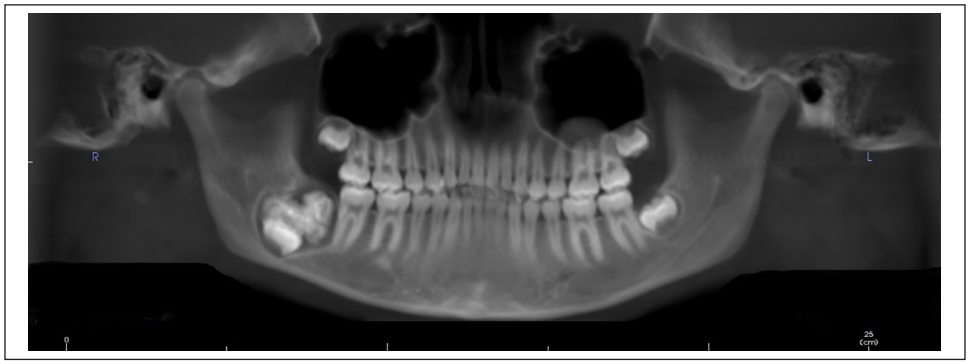

Figure 1. Well-circumscribed mixed radio-opaque/radiolucent mass near the mandibular canal in association with the developing crown of the right mandibular third molar.

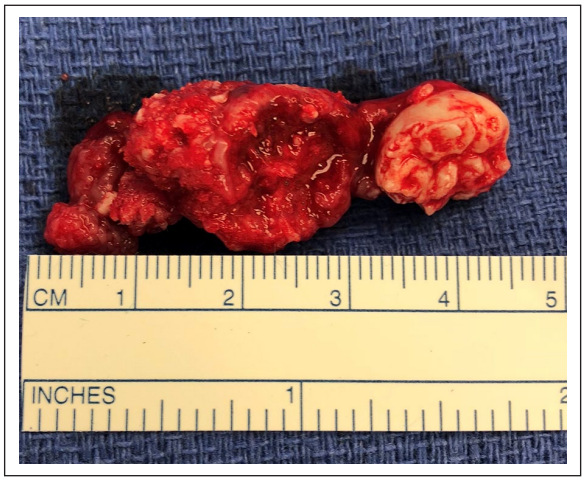

Figure 2. Mass in association with developing tooth.

chin and lower lip. Radiographic studies showed normal condyles and a well-circumscribed mixed radio-opaque/radiolucent mass near the mandibular canal in association with the developing crown of the right mandibular third molar (Figure 1). The patient underwent an enucleation and curettage of the hard right posterior mandibular lesion and extraction of the involved tooth (Figure 2). The inferior alveolar canal appeared to be intact, and the inferior alveolar nerve did not show clear involvement with the mass. However, involvement of dental and interdental branches of inferior alveolar nerve might account for this patient's paresthesia since the anatomy of inferior alveolar nerve may be quite complex, and the smaller nerve branches may not be observed radiographically. Microscopic examination of the specimen showed small islands and narrow cords of odontogenic epithelium, consisting of stellate cells in the center and palisading columnar cells in the periphery, located within a rich mesenchyme that contained round, stellate, or spindle-shaped cells (Figure 3), similar to those seen in the dental papilla. Cytologic atypia or mitotic figures were not observed. Enamel, dentin matrix, and foci of cementum formation were present adjacent to the epithelial structures (Figure 4). With these histopathologic findings, a diagnosis of AFO was rendered.

Discussion

AFO is a benign odontogenic tumor associated with a good prognosis.[7] The average age at presentation is 6 years in males and 9 years in females with a slight male predilection.[2] Kirjavainen et al.[8] found 60% of AFO cases located in the posterior region of the mandible and 94 out of 106 cases associated with the crown of an unerupted tooth. Furthermore, they described that the risk of recurrence following surgical removal was very low. Radiologically, AFOs are usually well-defined lesions with radio-opaque and radiolucent components. Sometimes, these lesions may contain a large amount of mineralized tissue resembling an odontoma.[2]

AFO belongs to a category of lesions referred to as benign mixed epithelial and mesenchymal odontogenic tumors, which include AFs, AFOs, and odontomas.[9] These lesions are neoplasms consisting of proliferating odontogenic epithelium in a background of cellular ectomesenchymal tissue that resembles the dental papilla, with varying degrees of inductive changes and dental hard tissue formation.[2] There is ongoing debate and disagreement among pathologists as to whether AFs and AFOs are neoplasms or are lesions at different stages in the development of odontomas. Some believe in the "maturation theory" suggesting that an AF will develop through a continuum of differentiation and maturation into an AFO and later into an odontoma.[5] Some authors claim that an AFO differs significantly from a hamartomatous odontoma by having a greater potential for causing bone deformity and destruction.[10,11] Trodahl[12] pointed out that odontomas go through a non-calcified stage of development that shows the histopathologic features of AF. Gardner[13] claimed that some lesions with the histopathologic appearance of an AFO are probably developing odontomas and that the histopathologic appearance of an AFO may be indistinguishable from developing odontomas, and an assessment of the coexistent clinical and radiological features may be of assistance in making a diagnostic distinction. In 2017, the WHO appeared to adopt

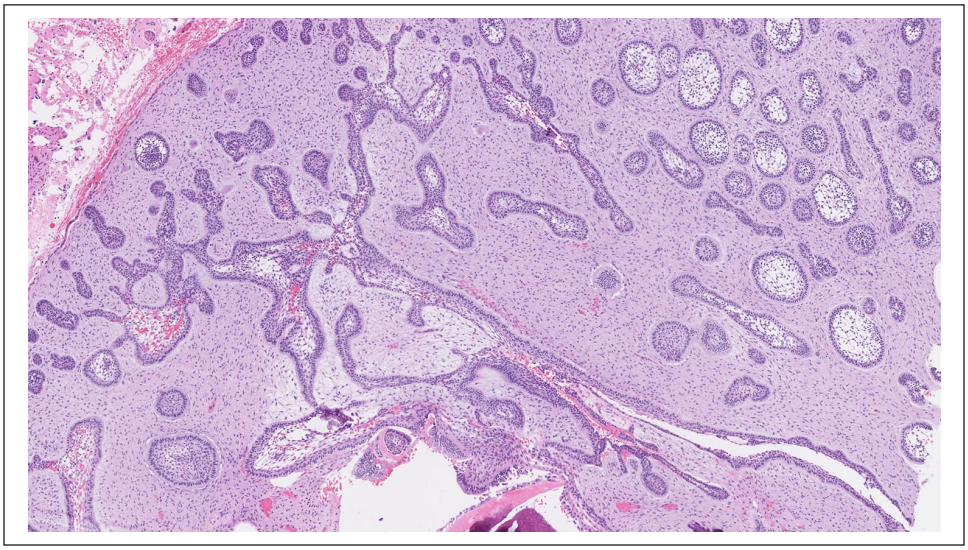

Figure 3. Cords and islands of ameloblastic epithelium within primitive ectomesenchymal tissue (hematoxylin-eosin, 20×).

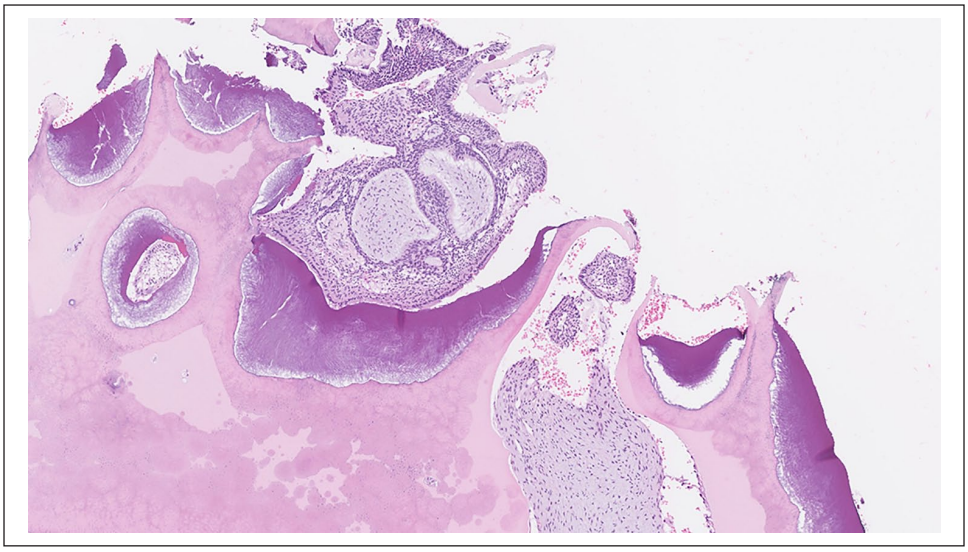

Figure 4. Odontogenic epithelium with the formation of dental hard tissue (hematoxylin-eosin, 40×).

a view similar to that of Gardner with respect to AFOs and developing odontomas.[14] A diagnosis of AFO was preferred in the case described herein due to the proportion of odontogenic epithelium observed. Smaller AFOs are usually treated with curettage and an attempt to preserve the involved tooth if the tooth does not interfere with the completion of the procedure. Larger AFOs can usually be enucleated with conservation of basal cortical bone.[2]

Conclusion

An AFO is a benign odontogenic tumor that shares a number of clinical and radiologic features with an AF and an odontoma and may in fact represent a stage in the development of an odontoma. Treatment with enucleation and curettage is usually successful. This treatment provided a desirable outcome in this case that was uniquely accompanied by preoperative paresthesia. Postoperatively, the patient was well with no evidence of residual/recurrent tumor/paresthesia.

Declaration of conflicting interests

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Ethical approval

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Informed consent

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