Ameloblastic Fibro-Odontoma in an Adolescent: A Case Report and Review of Literature

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Ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic tumor with the histologic features of ameloblastic fibroma (AF) but also contains enamel and dentin. It is most commonly observed in the pediatric population. Distinction between AFO and AF becomes important as ameloblastic fibromas are associated with higher recurrence rates of up to 18%, and 35% of these recurrent lesions can undergo malignant transformation to ameloblastic fibrosarcoma. Hence, for amelobastic fibroma, conservative curettage is recommended for the initial lesion and marginal resection is considered for recurrent cases. In contrast, AFO can be treated with simple curettage and the recurrence rate is approximately seven percent. Malignant transformation of AFO is exceedingly rare. Therefore, the treatment and prognosis differs for these two histologically similar neoplasms. We present a case of a 17-year-old boy who was initially diagnosed with ameloblastic fibroma upon biopsy, with subsequent curettage specimen showing AFO, which carries a better prognosis.

Key words: Ameloblastic fibro-odontoma, Ameloblastic fibroma, odontogenic neoplasm pediatric tumors, benign jaw lesions.

INTRODUCTION

Ameloblastic fibro-odontoma (AFO) is a rare benign mixed odontogenic tumor in which both the epithelial and ectomesenchymal components are considered neoplastic.[1] According to the most recent World Health Organization (WHO) classification, AFO shares similar histology with ameloblastic fibroma (AF), a benign neoplasm arising from odontogenic ectomesenchyme and odontogenic epithelium.[2] Unlike AF, however, AFO also contains enamel and dentin.[1,2]

AFO is seen most commonly in children, with an average age of diagnosis of ten years. A slight male predilection has been reported.[1,2] The posterior mandible is the most commonly affected location, and the lesion tends to present as either a unilocular radiolucent to mixed lesion. The degree of radiopacity observed is dependent on the amount of mineralized product produced by the tumor. While AF can exhibit progressive growth and result in significant deformity and bone destruction, AFO often produces no clinical symptoms.[1,3,4]

Herein, we report a case of a 17-year-old boy who was initially diagnosed with ameloblastic fibroma upon biopsy, with subsequent curettage specimen showing AFO, which carries a better prognosis. In addition, we review the relevant clinico-pathologic features regarding this entity.

Case Report

A 17-year-old boy was referred to an oral surgeon for evaluation of removal of his permanent third molars. The patient had no significant medical history or contraindications to routine dental care. His clinical examination was unremarkable. No soft tissue swellings or abnormalities were detected and no grossly carious lesions were present.

A panoramic radiograph was taken which demonstrated a large radiolucent lesion that appeared to be multilocular with areas of faint opacification. However, it was not clear if this was a mixed radiopaque/lucent lesion or if the opaque areas were radiographic artifacts. The lesion extended from the mesial root of the permanent mandibular left third molar to the mesial root of the permanent mandibular left first molar (Figure 1). No root resorption or

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divergence of the teeth was identified, and no boney expansion was evident. The patient denied any pain or paresthesia in the area. None of the teeth in the mandibular left quadrant were tender to percussion. A biopsy of the lesion was performed at the time of wisdom tooth extraction. The microscopic examination showed long, narrow cords of odontogenic epithelium supported by cellular primitive connective tissue stroma, diagnostic of ameloblastic fibroma (Figure 2).

With the working diagnosis of 'ameloblastic fibroma', a complete curettage of the lesion was performed. The second molar associated with the lesion could not be salvaged and was removed together with the tumor. Microscopic examination revealed small nests and long narrow cords of odontogenic epithelium within the cellular and primitive appearing stroma. In addition, there were some calcified structures consisting of foci of dentinoid and osteocementum-like materials in close relationship to the epithelial structures (Figures 3 and 4). These findings were consistent with a diagnosis of ameloblastic fibro-odontoma, which carries a better prognosis compared to ameloblastic fibroma.

Figure 1: Panoramic radiograph demonstrating a multilocular radiolucency with focal opacifications of the left posterior mandible, extending from the permanent mandibular left third molar to the permanent mandibular left first molar. No evidence of root resorption or divergence is seen.

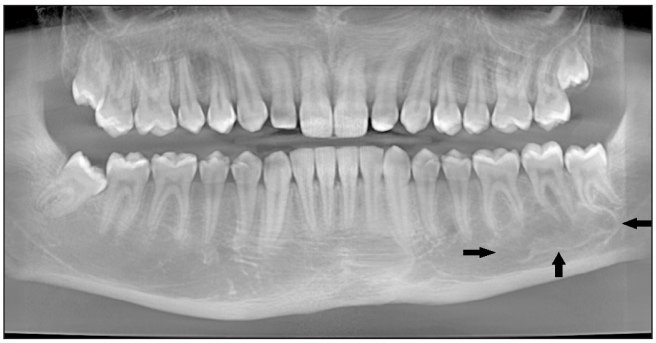
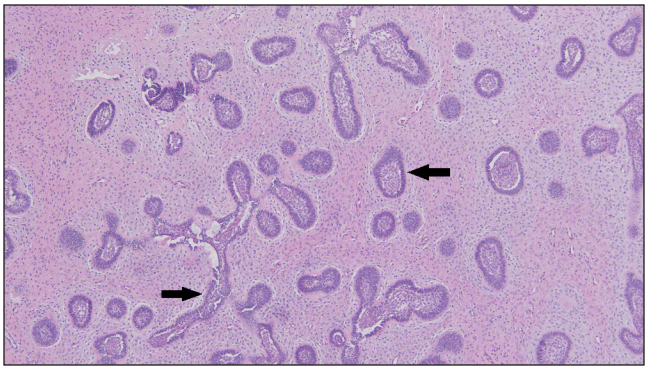


Figure 2: Photomicrograph from the biopsy specimen demonstrating small nests and long narrow cords of odontogenic epithelium within the cellular and primitive appearing stroma. As no dental hard tissues were seen, this lesion was initially diagnosed as an ameloblastic fibroma, H&E, x20.



DISCUSSION

Ameloblastic fibro-odontoma (AFO) is a benign mixed odontogenic tumor which typically presents in childhood. Similar to the ameloblastic fibroma (AF), it is composed of nests and cords of odontogenic epithelium within a background of cellular, primitive appearing stroma.[2] AFO is distinguished from AF by the presence of dentin and enamel within the ectomesenchyme.[1,2] These dental hard tissues often form adjacent to the epithelial structures. While the exact mechanism for their formation is not entirely understood, their close proximity to odontogenic epithelium suggests that the epithelium induces nearby mesenchymal cells to produce dentin-like material. This, in turn, may stimulate the odontogenic epithelium to form an enamel matrix-like substance.[4-6]

AFO is most commonly diagnosed in children ages eight to twelve, with a mean age of diagnosis of ten years.[2] AF is also most frequently seen in this age group. As was the case with our patient, AFO usually produces no clinical symptoms, but is rather discovered incidentally. Often, initial diagnosis is made when a radiograph is taken to determine why a tooth has failed to erupt.[1] Like many

Figure 3: Photomicrograph demonstrating nests and cords of odontogenic epithelium within a cellular, primitive appearing stroma. Foci of dentinoid and osteocementum-like materials are seen in close relationship to the epithelial structures, H&E x20.

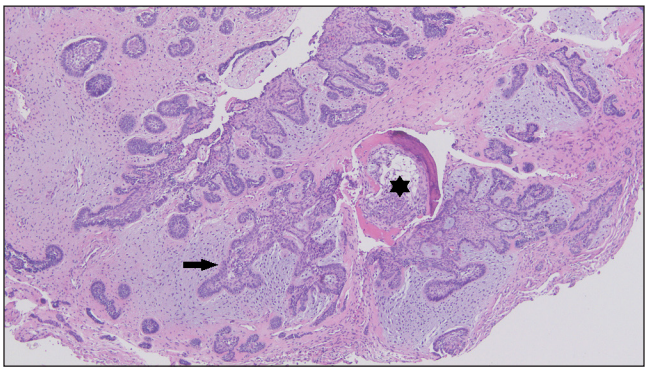
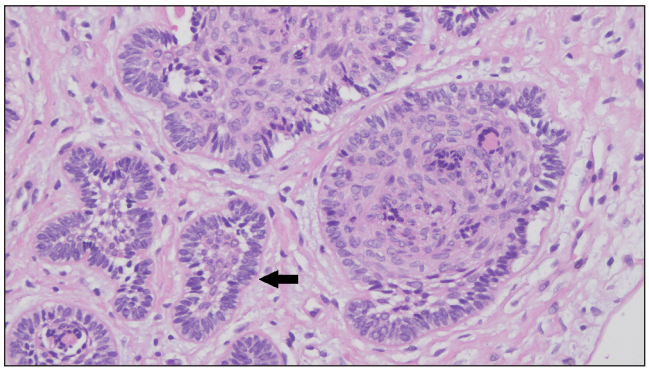


Figure 4: On high power magnification, the nests and cords of odontogenic epithelium demonstrate peripheral nuclear palisading, H&E, x100.



other odontogenic neoplasms, both AFO and AF have a predilection for the posterior mandible.[3]

Distinguishing between AFO and AF is important because ameloblastic fibromas are associated with higher recurrence rates of up to 18%, and 35% of these recurrent lesions can undergo malignant transformation to ameloblastic fibrosarcoma.[1,7,8] Therefore, the treatment and prognosis differs for these two histologically similar neoplasms. For amelobastic fibroma, conservative curettage is recommended for the initial lesion and marginal resection is considered for recurrent cases.[7,8] In contrast, AFO can be treated with simple curettage, and the recurrence rate is only approximately seven percent.[1] Malignant transformation of AFO is exceedingly rare. Howell and Burke report two cases of transformation of AFO to an ameloblastic fibrosarcoma, however they state that "this potential transformation alone does not justify radical treatment of all these benign lesions."[9] Furthermore, some authors believe that AFO is merely a developmental phase of a maturing odontoma, and do not advocate for treatment unless the lesion is interfering with proper tooth eruption.[10-12]

AFO tends to present as a well-defined unilocular radiolucency, although some larger lesions may be multilocular. An unerupted tooth is often present at the margin of the lesion; sometimes the crown of the tooth may be incorporated into the radiolucent defect. Prior to production of calcified materials, AFO will be completely radiolucent. As enamel and dentin are produced by neoplastic mesenchymal cells, radiopaque foci will develop on the radiograph, and present as a mixed radiolucent/opaque lesion. Hence both unilocular and multilocular radiolucent and mixed lesions of the jaw should be considered in the radiographic differential diagnosis.

The radiographic differential diagnosis of AFO is vast and can include both odontogenic and osseous lesions depending on the specific radiographic appearance and location of the tumor. The presence of opacifications may help exclude more common entities such as dentigerous cysts and keratocystic odontogenic tumors, which are wholly radiolucent. Adenomatoid odontogenic tumor (AOT) is a benign odontogenic neoplasm which is also seen in younger individuals and may present with radiographic opacifications (so-called "snow flake" opacities). Unlike AFO, however, AOT has a predilection for the anterior maxilla, where it is most often associated with an unerupted canine. Furthermore, the radiographic appearance of AOT is distinctive in that the lucency often extends to the apex of the involved tooth.[13] Calcifying epithelial odontogenic tumor (CEOT), also known as "Pindborg" tumor, tends to occur in the posterior mandible and often has "driven-snow" calcifications. CEOT should not be confused with AFO, however, as the former is an exceedingly rare tumor found primarily in older adults.[14] An odontoameloblastoma (OA) is another rare mixed odontogenic tumor which may radiographically mimic an AFO. However, OA has a behavior profile similar to a conventional ameloblastoma, and will usually present with clinical symptoms of root resorption, jaw pain, and boney expansion.[15]

Distinction between AF and AFO is made by the radiographic lack of opacifications in the former as well as the histologic differences previously discussed.[2] An AFO with a large amount of mineralized product may be confused for a benign fibro-osseous lesion, such as a juvenile variant of ossifying fibroma. In the case of ossifying fibroma, however, the patient will often present with boney expansion which is not typically seen in patients with AFO.[16]

CONCLUSIONS

AFO is a rare yet well-defined lesion characterized by neoplastic changes in both odontogenic epithelium and ectomesenchyme. In addition, the tumor produces odontogenic hard tissues (dentin and enamel) which is seen in close proximity to the epithelial component of the lesion. As was the case with our patient, AFO often produces no clinical symptoms, but rather is discovered incidentally when dental radiographs are taken for other clinical purposes. Distinguishing between AF and AFO is essential, as these two histologically similar neoplasms are managed differently due to the higher recurrence rate and risk of malignant transformation of the former, hence requiring frequent follow-up for AF. One must also include other similar appearing odontogenic and osseous lesions when establishing a radiographic differential diagnosis for this entity.

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