Ameloblastic fibro-odontoma: a case report

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Ameloblastic fibro-odontoma (AFO) is a rare mixed odontogenic tumor consisting of both ectodermal and mesenchymal components. It occurs predominantly in children and adolescents, especially in the mandibular posterior region. Histopathologically, AFO exhibits the combination of ameloblastic fibroma-like tissue and complex odontoma. A case of AFO in a 1-year-old child was presented. It produced an expansile lesion over the upper right anterior region and showed typical histopathological features of AFO. She was treated by enucleation with no recurrence observed after a follow-up period of 1 year.
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INTRODUCTION

Ameloblastic fibro-odontoma (AFO) is a rare mixed odontogenic tumor consisting of both ectodermal and mesenchymal components. According to WHO classification of odontogenic tumors, AFO is defined as a lesion similar to ameloblastic fibroma, but shows inductive changes that lead to the formation of dentine as well as enamel.[1] AFO generally occurs in young patients within the first two decades of life. It is rarely encountered in adults.[2-9] It has a predilection for male patients.[2,4,5,8,10]

The majority of AFOs occur in the posterior region of the jaws, especially the mandibular posterior region.[2,7,8,11] AFO is an asymptomatic, slowly growing and expansile lesion, the majority of which are associated with unerupted teeth.[5,7,9,12,13] The failure of tooth to erupt usually bring this lesion to medical attention.[7,8] Radiographically, AFO shows a well-circumscribed unilocular or, on rare occasion, multilocular radiolucency containing variable amounts of radiopaque material.[7-9,12] This radiopaque material usually has radio-density comparable to that of tooth structure.[8] AFO sometimes contains only a minute amounts of radiopaque material rendering it translucent radiographically.[7] AFO consists of 3 components 1) ectodermal part characterized by odontogenic epithelium 2) immature mesenchymal tissue resembling dental pulp 3) mineralized part made up of irregularly shaped dental structure.[5,14] The treatment of choice for AFO is conservative surgery by enucleation along with the removal of associated unerupted tooth.[8-12,15] It is easily removed from the bony bed, since it is well-circumscribed and possesses little propensity to invade surrounding bone.[9,10,15]

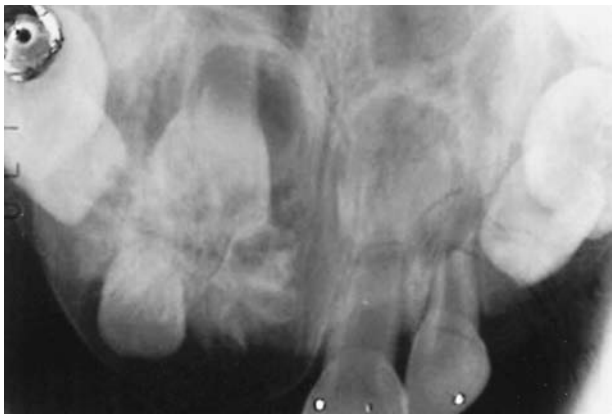

Figure 1. Radiograph showing a well-circumscribed unilocular radiolucency containing irregular-shaped radiopaque material as well as teeth #51 and 52.

CASE REPORT

A 1-year-old female child was brought to Sawanpracharak Hospital with the chief complaint of gingival enlargement and missing upper right anterior teeth. Her mother noticed a rapid gingival enlargement at the upper right anterior arch. Intraoral examination revealed a bucco-lingual expansion at the area of upper right anterior teeth covered by intact mucosa. The teeth #61 and 62 erupted normally, while the teeth #51 and 52 were not seen in the oral cavity. The radiograph showed a well-circumscribed unilocular radiolucency containing irregular-shaped radiopaque material as well as

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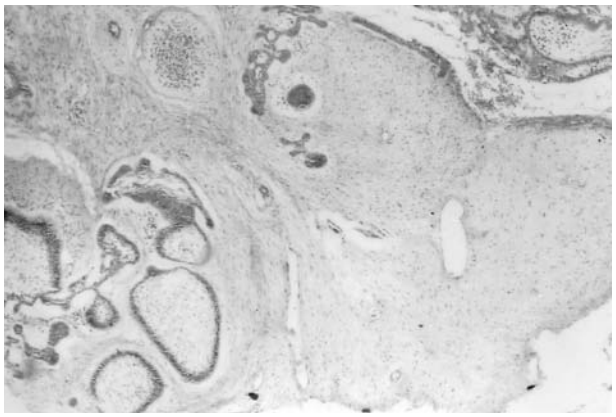

Figure 2. Photomicrograph showing islands of odontogenic epithelium. These islands consisted of loosely arranged central area resembling stellate reticulum surrounded by a layer of ameloblast-like columnar cells. (Hematoxylin and eosin stain, original magnification)

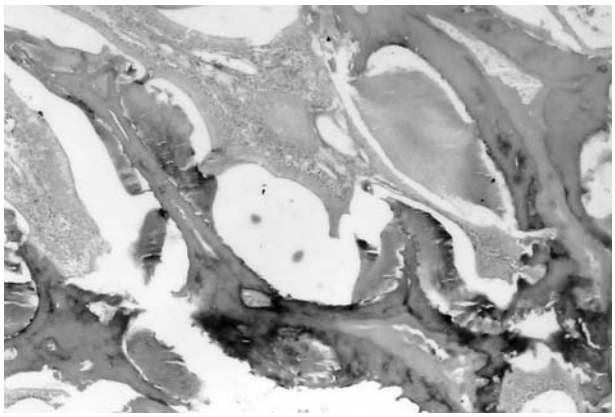

Figure 3. Photomicrograph showing calcified materials: enamel matrix, dentine and cementum, arranged in haphazard pattern without any relation to normal anatomical orientation. (Hematoxylin and eosin stain, original magnification)

teeth #51 and 52 (Figure 1). The clinical diagnosis was odontoma. The patient was treated by enucleation of the lesion as well as teeth #51 and 52 under general anesthesia. The post-operative course was uneventful.

The pathological examination of the specimen revealed a pale pink, round-shaped soft tissue sized 1.5x1.5x1.0 cm^3. The lesion showed islands and strands of odontogenic epithelium surrounded by embryonic cellular connective tissue. The epithelial component formed islands or strands that consisted of loosely arranged central area resembling stellate reticulum surrounded by a layer of ameloblast-like columnar or cuboidal cells. The mesenchymal component was primitive connective tissue consisting of delicate collagenous fibers interspersed with fibroblasts reminiscent of dental papilla of the tooth germ. The mineralized materials consisted of irregular masses of various shapes and sizes of dentine, enamel matrix and cementum with entrapped odontogenic epithelium arranged in a haphazard pattern without any relation to normal anatomical orientation.

The one-year follow-up revealed no recurrence at the operated site and tooth bud of #11 and 12 developed normally.

DISCUSSION

This case was diagnosed as AFO because it fulfilled the histopathological criteria for the diagnosis of AFO. The interesting findings of the present case compared to previously reported AFO cases are an extremely young age, the upper anterior position and the rapid growth of the present case.

AFO should be differentiated from AF and ameloblastic fibrodentinoma. AF occurs at a higher age than AFO.[8,16] Histopathologically, AFO demonstrates the product of inductive changes, dentine as well as enamel, in addition to ameloblastic fibroma-like tissue. AFO advances from ameloblastic fibrodentinoma in that it elaborates not only dentine, but enamel as well.[1]

AFO should also be differentiated from odontoameloblastoma, since there are differences between these entities in both histopathological features and clinical behavior. The histopathological features of AFO are as previously described. They are in contrast to odontoameloblastoma in which soft tissue element is mainly composed of proliferating odontogenic epithelium resembling ameloblastoma. AFO behaves in a less aggressive manner as AF,[9] while odontoameloblastoma behaves in a more aggressive and invasive manner of classic ameloblastoma.[1,9]

AFO may be confused with lesions belonging to the fibro-osseous group due to the mixed radiolucent-radiopaque radiograph, but this problem can be easily resolved through microscopic examination because of the clearly different histopathological features. Although AFO may appear on the differential diagnosis of mixed radiolucent-radiopaque lesion in young patients, most of them are clinically diagnosed as odontoma. Only after histopathological examination does it become the definite diagnosis.

The most challenging task is to differentiate AFO from developing odontoma. Developing odontoma may have the histopathological appearance of AFO. Consequently, the clinical findings are important in differentiating between the two lesions. A small lesion located over an unerupted molar in a child is more likely to be a developing odontoma, while an expansile lesion of the jaws is more likely to be an AFO.[6,7]

Confusion exists regarding the relationship between AF, AFO, and odontoma. Some consider them as separate entities.[17,18] Others regard them as chronological stages in a continuum starting from AF at one end and odontoma at the other end with AFO in an intermediate

stage.[19,20] The result from the analysis of 33 mixed odontogenic tumors by Slootweg[2] contradicted the latter notion, since he found that the mean age of the patients with AFO was lower than that of the patients with the more primitive looking AF. If the latter notion held true, the mean age of the patients with AFO, which is supposed to subsequently develop from AF would be higher than that of the patients with AF. The age of our patient adds further contradictory evidence to the latter notion since our patient is an extremely young child.

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