Adenomatoid odontogenic tumor associated with dilacerated root: An unusual case report

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Abstract

Adenomatoid odontogenic tumor (AOT) is a rare benign neoplasm of epithelial origin. AOT occurs mainly in the second decade of life, and the most common location is anterior maxilla. It rarely affects the mandible. Most of the AOTs may be located intraosseously but few have been reported to occur within gingival structures. AOTs located intraosseously may be seen associated with unerupted tooth (follicular variant) or may not (extra-follicular variant) be associated with unerupted tooth. Here, we report a rare case of presumably follicular variant of AOT associated with mandibular canine that presented as an extra-follicular variant leading to root dilaceration of the mandibular canine after eruption of the tooth.

Keywords: Adenomatoid, extra-follicular, follicular, odontogenic, tumor

Introduction

In 1969, Philipsen and Birn proposed the widely accepted and the currently used name adenomatoid odontogenic tumor (AOT), which was included in the first classification of odontogenic tumor by histologic typing of World Health Organization (WHO) in 1971. In 2005, WHO had recategorized AOT as benign odontogenic tumor of epithelial origin with mature, fibrous stroma without Odontogenic ectomesenchyme. Some of the investigators even believe it to be a hamartoma. AOT is an uncommon benign odontogenic tumor occurring with a relative frequency of 2.2-7.1%. It exists as three clinical variants: Follicular (related to unerupted tooth), extra-follicular (not related to unerupted tooth), and peripheral (attached to the gingival structures). AOT is referred to as “2/3rd tumor” because 2/3rd of the cases occur in the maxilla, 2/3rd of the cases occur in young woman, 2/3rd of the cases occur in unerupted teeth and 2/3rd of the cases have been found to arise in relation to anterior tooth particularly canine.

Here we report a rare case of presumably follicular AOT in 15-year-old female which arose in association of left mandibular canine and later with eruption of the tooth presented as an extra-follicular variant leading to the root dilaceration of left mandibular canine.

Case Report

A 15-year-old female patient reported with the chief complaint of the painless swelling in the mandibular anterior region since 6 months. The swelling was not associated with any history of trauma, pain or discharge. On extra-oral examination, lymphadenopathy was not observed. On extra-oral examination, labial cortical plate expansion was seen in relation to tooth 33 with apparently normal overlying mucosa [Figure 1]. On palpation, the swelling was hard and non-tender. Electrometric pulp testing of tooth 33 suggested that the tooth was vital. Orthopantomogram showed 33 with dilacerated root and periapical radiolucency [Figure 2]. The provisional diagnosis of lateral periodontal cyst was made. On surgical exploration, there was the buccal cortical plate resorption and the lesion was totally enucleated. The gross examination of the specimen showed a single spherical greyish white soft tissue measuring 1 cm × 1.5 cm and cut surface revealed a solid grayish-white tumor mass with well-defined capsule [Figure 3]. Microscopy revealed multisized solid nodules of cuboidal and columnar epithelial cells forming nests or rosette-like structure with minimal stromal connective tissue and spindle shaped cells in the spaces between the epithelial nodules which are encapsulated with a thick,
inflamed fibrous capsule [Figure 4a]. Ameloblast-like epithelial cells forming duct-like structures, eosinophilic amorphous tumor droplets amongst the epithelial cells; leisegang ring formation and eosinophilic amorphous amyloid like deposits were observed [Figure 4b]. The final histopathological diagnosis of AOT was obtained. The patient was kept under regular follow-up. The patient has not shown any signs of recurrence in the 6 months follow-up period after surgery.

Discussion
AOT is a rare benign odontogenic neoplasm with predilection for occurrence in females and occurs mainly in the second decade of life as seen in our case.[2,6-8] The lesions usually present as asymptomatic jaw swelling and are relatively small in size not exceeding 1-3 cm in diameter, as seen in this case.[8]

AOT exists as three clinical subtypes: follicular type (in 73% of AOT) which is intraosseous in location and is associated with an unerupted tooth (usually canine); the extra-follicular variant (24%) which is located intraosseously but is not associated with unerupted tooth, and the peripheral form which is the most rare (3%) and occurs within gingival mucosa.[6,9]

On radiographs, the intraosseous follicular variant of AOT shows a well-delineated, unilocular radiolucency surrounding the crown of a retained tooth, a picture indistinguishable from dentigerous cysts. Minute radiopacities around the retained tooth may be found in AOT and are considered a characteristic but not pathognomonic finding. About two out of every three AOT show distinct radiopaque calcification on radiographs.[6,9]

The extra-follicular variant presents as a unilocular, well-defined radiolucency found between, above, or superimposed on the roots of erupted teeth and often resembles a cystic lesion.[6]

Our case presented as an extra-follicular AOT that was diagnosed clinically as a cystic lesion.

AOT affects more commonly the maxilla than the mandible in a ratio of 2.1:1.[10] The case reported here is considered uncommon as it involved the mandible. Our case is also unique as it is associated with dilacerated tooth and root dilaceration within

![Figure 1: Intraoral photograph showing labial cortical plate expansion on the mandibular anterior alveolus in relation to teeth 33](image1)

![Figure 2: Orthopantomogram showing periapical radiolucency in relation to dilacerated tooth 33](image2)

![Figure 3: Cut surface of the gross specimen that revealed a solid grayish-white tumor mass with well-defined capsule](image3)

![Figure 4: (a) Multisized solid nodules forming and amyloid like eosinophilic deposits that are encapsulated with a thick, regular inflamed fibrous connective tissue (×10). (b): Solid nodules of cuboidal and columnar epithelial cells forming nests and rosette-like structure along leisegang ring formation and spindle-shaped cells in the spaces between the epithelial nodules (×40)](image4)
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...the lesion are only infrequently reported in the literature.[11] We hypothesize that the AOT in this patient evolved as a follicular variant associated with 33. After the eruption of 33, there could be shift of position of the lesion, which impeded and changed the pathway of development of the root resulting in root dilaceration.

Considerable amount of debate is going on regarding the hamartomatous, neoplastic or cystic nature of AOT.[7] It was considered to be a hamartoma clinically and histologically, due to small size of the tumor at the time of diagnosis and the lack of recurrences after removal. However, it had also been described as a slow growing neoplasm that is detected at an early stage, before it reached a clinically noticeable size.[7,11]

Amyloid like material resembling that of calcifying epithelial odontogenic tumor (CEOT) are also present and the presence of which has led some workers to propose the existence of combined AOT and CEOT. However, CEOT like areas were considered to be within normal histopathological spectrum of AOT.[12] The calcified materials seen in AOT have been considered to be an abortive form of enamel, dentin, enamel and dentin, cementum, dentin, and cementum or dystrophic calcifications, but their exact nature remains a controversy.[13]

Owning to its benign behavior, slow growth and clear delimitation, as well as its low tendency to recur, the treatment of choice for AOTs is conservative surgical excision.[14]

Conclusion

We conclude that the reported case presumably evolved as follicular variant that later presented as extra-follicular variant resulting in the dilacerations of associated erupted tooth.

References