Adenomatoid Odontogenic Tumor of Anterior Maxilla; A Case Report

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Abstract
Here, we report a case of adenomatoid odontogenic tumor in anterior maxilla in a young girl aged 14 years. Adenomatoid odontogenic tumor is a rare, benign, and distinct slow-growing odontogenic neoplasm which results in painless expansion of jaws. It usually occurs in the anterior maxilla followed by posterior maxilla and rarely in the mandible with an age range of 3 to 82 years and a relative frequency of 2.2% to 7.1% and showing a female predilection (1.9:1).

Keywords
Adenomatoid odontogenic tumor, benign oral tumor, anterior maxillary tumor, hamartoma

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Introduction
Of all odontogenic tumors, adenomatoid odontogenic tumor represents 3% to 7%. It exhibits duct-like structures and tubular characteristics microscopically similar to ameloblastoma which led to this lesion being designated as “adenoameloblastoma.”1 “WHO classification of odontogenic tumours in 2005 classified adenomatoid odontogenic tumour into the first group of tumors (odontogenic epithelium without ectomesenchyme”).2 Stafne in 1948 first identified it as a benign lesion originating from the odontogenic epithelium and stated that it is nonaggressive and slow growing in nature. Some authors have even classified these lesions as hamartomas.3 Its distinctive clinicopathologic profile although not pathognomonic is considerably different among other odontogenic tumors because most adenomatoid odontogenic tumors occur in association with an unerupted maxillary cuspid in teenage girls.4 Adenomatoid odontogenic tumor appears in 3 clinical variants: follicular, extrafollicular, and peripheral type.5 Here, a case of adenomatoid odontogenic tumor is presented highlighting the importance of accuracy of diagnosis of such lesions for its efficient management.

Case Report
A 14-year-old female patient reported with a chief complaint of painless swelling with occasional pus discharge in her anterior maxillary region for the past 6 months. The lesion was not associated with any history of trauma, pain, and lymphadenopathy. The skin over the lesion and the surrounding area appeared normal and the margins were diffuse. On palpation, there was no local rise in temperature, no tenderness, and no discharge.

Intraoral examination revealed swelling of size approx 4 cm × 3 cm obliterating the left side labial vestibule extending from maxillary lateral incisor to the second molar medio-laterally. Superoinferiorly, it extended from middle third of labial aspect of edentulous area in relation to 24,25 to the depth of labial vestibule. The swelling was firm in consistency. On palpation, swelling was nontender, nonfluctuant, nonpulsatile, noncompressible, and nonreducible with no discharge (Figure 1). Patient gave a history of surgical dental extraction 6 years back in the same region. Cone beam...
computed tomography was advised which revealed a round, well-circumscribed, radiolucent lesion with well-defined distinct radiopaque margins 4 cm × 3 cm in size extending from left maxillary lateral incisor to the maxillary second molar extending into the maxillary antrum (Figure 2). Based on the history of extraction, clinical and radiographic findings, a provisional diagnosis of adenomatoid odontogenic tumor was made. Aspiration of the lesion was done, which was negative.

Incisional biopsy of the lesion was done under local anesthesia. The sections revealed fibrocollagenous tissue lined focally by a stratified squamous epithelium with a superficial nodular proliferation of round to polygonal cells in solid sheets and duct-like structures and rosettes suggestive of odontogenic epithelial cells. Eosinophilic fibrillar material was present between tumor cells and within duct-like structures, also focal calcification was noted. Based on these findings, a histopathologic diagnosis of adenomatoid odontogenic tumor was made (Figure 3). Enucleation of the tumor under general anesthesia was chosen as the treatment option.

After inducing general anesthesia and intraoral site preparation, a crevicular incision was placed extending from the left maxillary lateral incisor to the left maxillary second molar and a trapezoidal flap was raised followed by subperiosteal elevation of the flap to expose the tumor mass. The labial cortex was found to be resorbed. Careful excision of the lesion along with its capsule was done in toto. Gross examination of the specimen showed a single spherical-shaped soft tissue mass measuring approximately 3 cm × 3 cm (Figure 4).

The surgical specimen (Figure 5) was sent for histopathologic examination with a confirmatory diagnosis of adenomatoid odontogenic tumor. Regular clinical follow-up of the patient was done for a period of 28 months and no signs of recurrence were observed.
Discussion

The first identifiable case of adenomatoid odontogenic tumor is difficult to pin down because this lesion has been given numerous names, the earliest reported case was from Norway by Harbitz in 1915. Stafne reported the first series of adenomatoid odontogenic tumor in 1948 under the title “epithelial tumours other lesions) which is well demarcated with smooth cortical border. Displacement of neighboring teeth is a more common finding compared to root resorptions. Approximately, 78% of adenomatoid odontogenic tumors show calcified deposits and are associated with the crown of an unerupted permanent tooth in 71% of cases.

Gross examination of majority of tumors reveals a soft, roughly spherical mass with a veritable fibrous capsule. Upon sectioning, the tumor may exhibit white to tan, solid to crumbly tissue, or one or more cystic spaces of varying size; minimal yellow brown fluid to semisolid material; fine, hard “gritty” granular material; and one to several larger calcified masses. Additionally, intact dentigerous specimens demonstrate the crown of a tooth embedded in the solid tumor mass or projecting into a cystic cavity.

The tumor is made up of a cellular multinodular proliferation of spindle, cuboidal, and columnar cells in a variety of patterns; usually scattered duct-like structures, eosinophilic material, and calcifications in several forms and a fibrous capsule of variable thickness.

Due to uniformly benign biologic behavior of nearly all typical adenomatoid odontogenic tumors and consistent presence of a well-developed fibrous capsule, complete surgical excision—usually accomplished by enucleation and curettage—is the treatment of choice (excision in toto).

Adenomatoid odontogenic tumor is referred to as “slow” or “very-slowly growing” but no report of measurements of growth rate over a course of time could be located. The large size of a few of the reported cases in young children from underdeveloped countries indicate that some cases have shown at least a moderate rate of growth. Recurrence is exceptionally rare of these lesions.

Conclusion

Adenomatoid odontogenic tumor is an uncommon odontogenic lesion, but it can be usually identified from its clinical and radiographic appearance. The clinico-radiographic profile of adenomatoid odontogenic tumor observed in this study agrees with that commonly reported in literature. It is important to correctly distinguish and diagnose this lesion to eliminate the risk of unnecessarily mutilating surgery for patients. Conservative surgical enucleation is the treatment of choice as the lesion is well encapsulated and can be separated from the bone easily. Also, its recurrence is very rare.

Declaration of Conflicting Interests

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Statement of Informed Consent and Ethical Approval

Necessary ethical clearances and informed consent was received and obtained respectively before initiating the study from all participants.

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