ADENOMATOID ODONTOGENIC TUMOUR – AN UNUSUAL EXTRAFOILLCULAR VARIANT OF THE POSTERIOR MANDIBLE

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ABSTRACT
Adenomatoid odontogenic tumor (AOT) is a relatively uncommon distinct odontogenic neoplasm. Although this lesion was formerly considered to be the variant of ameloblastoma, its clinical features and biologic behavior indicate, that it is a separate entity. It is a tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive change in the stroma. It is a benign and slow growing tumor which is usually located in the anterior region of the maxilla without pain, and represents 3% of all odontogenic tumors. Most adenomatoid odontogenic tumors (AOTs) occur intra-osseously. They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship, whereas the extrafollicular type has no relation with an impacted tooth, and the peripheral variant is attached to the gingival structures. The aim of this paper is to present a rare case of an extrafollicular AOT occurring in the posterior aspect of the mandible which is distinct.

KEY WORDS: Adenomatoid Odontogenic tumour, Intraosseous, Extra follicular.

INTRODUCTION
Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905. However, a variety of terms have been used to describe this tumor. In 1969 Philipsen and Birn proposed the term AOT, indicating that it did not constitute a variety of ameloblastoma, and was accepted as such in the first WHO classification of odontogenic tumors established in 1971. Although this lesion was formerly considered to be the variant of ameloblastoma, its clinical features and biologic behavior indicate that it is a separate entity. Many different names like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratomatous odontoma have been used before to define the lesion currently called AOT.

Most of the adenomatoid odontogenic tumors (AOTs) occur intra-ossesously. They surround the crowns and are attached to the necks of unerupted teeth in a true follicular relationship. Some, however have no association with unerupted teeth and a few occur on the gingiva in an extraosseous location. For the follicular variant there is histologic and immunohistochemical evidence that they originate from the reduced enamel epithelium (REE) of the dental follicle.

Case Report
An 18 year old female visited to the Department of Oral Medicine and Radiology, SVS Institute of Dental Sciences, Mahabubnagar, with a chief complaint of swelling on her left side of the face since six months, and pain in the lower left last tooth since three months. The swelling had a slow onset and gradually increased in size leading to disfigurement of the face. A broad review of systemic examination revealed neither any abnormalities nor presence of any relevant diseases.

Extra oral (Fig.1) inspection revealed facial asymmetry due to the presence of a solitary diffused swelling measuring 3.5x5.5 cm in size on the left side of the face. The swelling was extending from the zygomatic arch region superiorly to the inferior border of the mandible inferiorly. Anteriorly it was extending 2 cm. away from the left corner of the mouth to the posterior border of the ramus posteriorly i.e., almost covering the whole of the ramus region. On palpation the swelling was nontender, rubbery to hard in consistency and fixed to the underlying bone. No features of reducibility, compressibility, or any pulsations was noted.

On Intra oral examination (Fig.2), a solitary swelling noted in the left mandibular ramus and retromolar area was obliterating the vestibule.
Superiorly it was extending around 2 cm from the occlusal plane of the lower teeth, anteriorly it was extending from distal margin of lower left first molar to the retromolar area posteriorly. The colour of the swelling was normal mucosal colour. The margins were ill-defined and diffused. On palpation it has variable consistency from rubbery to hard in nature. There was grade-II mobility of the second molar tooth with mild amount of depressability in the socket was noted.

It was provisionally diagnosed as Ameloblastoma with the differential diagnosis of Ameloblastic fibroma and Odontogenic myxoma. Fine needle aspiration was found negative.

An Orthopantomograph (Fig.3) revealed an unilocular, roughly rectangular radiolucency of 4x5cm in the left ramus region with well defined sclerotic borders. An intraoral periapical radiograph revealed, missing third molar tooth, and the roots of the second molar tooth was mesially drifted with no signs of root resorption (Fig.4). Computed tomography revealed a hypodense area noted in the left ramus of the mandible with well-defined outline suggesting a benign and well capsulated, noninvasive, soft tissue tumour without any calcifications.(Fig.5, Fig.6 and Fig.7).

An incisional biopsy was made through the window created by the extraction of the second molar. Histopathology (Fig.8) revealed deeply placed islands of darkly staining cells with peripheral layer of columnar like cells. Some areas appear to form subtle duct like structures, with areas of coarse calcifications are also evident, features suggestive of Adenomatoid odontogenic tumour (AOT) concurrent with extra-follicular variant of AOT.

Discussion

Adenomatoid odontogenic tumor (AOT) is a tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive change in the stroma. It is an uncommon, benign and slow growing tumor which is usually located in the anterior region of the maxilla without pain, and represents 3% of all odontogenic tumors. It is largely limited to the younger patients, and two thirds of all cases are diagnosed when patients are 10 to 19 years of age. There is a slightly female over male incidence, an almost 2:1. The tumor is usually associated with an un-erupted, teeth, frequently canines or lateral incisors.

The lesions are typically asymptomatic, but may cause cortical expansion and displacement of the adjacent teeth, as in the case reported here. The slow growing nature of the lesion may cause the patients tolerate the swelling for years until it produces an obvious deformity. The origin of the AOT is controversial. Because of its predilection for tooth-bearing bone, it is thought to arise from odontogenic epithelium. Epithelial remnants from the dental lamina and the accessional dental lamina (sometimes together called the parent dental lamina) could be the likely source of origin of the peripheral variants and intraosseous variants of AOT confined to the permanent molar region.
The tumor has three clinicopathologic variants, namely- Intraosseous follicular, Intraosseous extrafollicular, and Peripheral. The follicular type (73% of all AOT cases) is associated with an unerupted tooth whereas extrafollicular type (24%) has no relation with an impacted tooth and the peripheral variant (3%) is attached to the gingival structures. Follicular and extrafollicular types are over two times more located in the maxilla than in the mandible and most of the tumors involve anterior aspect of the jaws. Remarkably, all variants of AOT show identical histology. The histological typing of the WHO defined the AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor is well encapsulated and shows an identical benign behavior. Radiographically, they usually appear unilocular, may contain fine calcifications, and irregular root resorption is rare. The usual radiographic appearance is a well defined corticated or sclerotic border. Occasionally the calcifications are small and with well-defined borders, like a cluster of small pebbles.}

The differential diagnosis can be made with ameloblastoma, dentigerous cyst, and ameloblastic fibro odontoma. The patient described in this report has no root resorption, but there is displacement of the adjacent teeth and also the tumor was not associated with an impacted tooth. Upon gross sectioning, the tumor exhibited 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. Remarkably, all variants of AOT show identical histology. The histological typing of the WHO defined the AOT as a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor is well encapsulated and shows an identical benign behavior. Enucleation and Currettage is the most common treatment modality for this tumor. Conservative surgical excision is adequate because the tumor is not locally invasive, is well encapsulated, and is separated easily from the bone. The recurrence rate is 0.2%.
CONCLUSION:

Although Adenomatoid Odontogenic tumours are common, the extra follicular variant of AOT in the posterior part of the ramus of the mandible is very rare. Though it is benign, its large sized tumour extent as in this case, has led to partial mandibulectomy, which would have been avoided if the tumour was detected in early days of the tumour onset.

References:
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