Case Report

A true neoplasm: Adenomatoid odontogenic tumor

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is relatively an uncommon oral tumor, which accounts for about 3-7% of all odontogenic tumors. As the histogenesis is still uncertain, it is sometimes categorized as a hamartomatous lesion rather than a true neoplasm. We report a case of AOT in the left maxillary anterior region in 23-year-old female patient. The tumor showed some unusual and aggressive features that suggested it was a true neoplasm.

Key words: Adenomatoid odontogenic tumor, neoplasm, odontogenic tumor

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a relatively uncommon distinct odontogenic neoplasm that was first described by Steensland in 1905.[1] However, number of terms have been used to describe this tumor. Unal *et al*. produced a list containing all nomenclatures for AOT reported in the literature.[2]

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.[3]

It was first reported by Harvetz in 1955 as cystic adamantoma. Phillipsen and Birn proposed the widely excepted and currently used name AOT, the term that was adopted by WHO classification of odontogenic tumor in 1971.[4]

Adenomatoid odontogenic tumor is a slow growing, benign, epithelial tumor that presents with clinical signs of the absence of teeth and areas of deformity in very large lesions. AOT is usually seen during the second and third decades. The incidence is higher in females than in males and accounts for 3-7% of all odontogenic tumors, AOT is usually located in the anterior region of the maxilla within which an impacted tooth exist.[5]

Tumor appears as intraoral-extraoral swelling in the maxilla, and it is sometimes referred to as 2/3rd tumor because it occurs in maxilla in about 2/3rd cases, about 2/3rd cases arises in young females, 2/3rd cases are associated with unerupted tooth, 2/3rd affected teeth are canine.[4]

In this report, a 23-year-old female patient had an AOT with a diameter of 15 cm × 5 cm located in the anterior region of the maxilla involving lateral incisor, canine, and premolar.

CASE REPORT

A 23-year-old patient reported to the Department of Oral and Maxillofacial Pathology with a chief complaint of swelling on the left side of the face since 1 year.

Initially, swelling was smaller in size that is near to 2 mm × 3 mm and gradually increased to present size (15 mm × 5 mm). Excision of tissue was done 2 years back by a private practitioner. Swelling recurred within a year to the present size.

Extraorally ovoid swelling was seen resulting in facial asymmetry. The swelling was irregular in shape

Access this article online	
Quick Response Code:	Website: www.ccij-online.org
	DOI: 10.4103/2278-0513.149046

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measuring 15 mm × 5 mm. On palpation, swelling was bony hard in consistency, nontender and immobile with no bleeding or pus discharge. It was smooth with well-defined margins. Intraorally swelling was present on attached gingiva on the left side, extending from the distal surface of lateral incisor to the distal surface of the first premolar, obliterating the buccal vestibule enclosing tooth 23 completely. Overlying mucosa was normal and stretched with no impacted tooth.

On radiographic examination, orthopantomogram showed a well-defined pear shaped radiolucency extending from radicular portion of 22 to the mesial aspect of 24 involving 23 [Figure 1]. Root resorption of 23 and 24 was evident. Maxillary occlusal radiograph reveals expansion of buccal cortical plates with root resorption of 23 [Figure 2].

Based on clinical and radiographic features, a provisional diagnosis of AOT was made. The radiographic findings of AOT frequently resemble other odontogenic lesions such as dentigerous cysts, calcifying odontogenic cysts, calcifying odontogenic tumors, globule-maxillary cysts, ameloblastomas, odontogenic keratocysts, and periapical disease.[6]

Surgical enucleation was performed, and the specimen was sent for the histopathological examination.

On gross examination, the tumor was grayish white in color, firm to soft in consistency, pear-shaped, measuring 15 mm × 5 mm × 3 mm. The teeth associated with lesion showing roots of 23, 24 were resorbed also extracted.

Under scanner view (×4) hematoxylin and eosin stained section revealed duct-like microcysts and convoluted structures with scanty stroma.

Under high power (×40) a well-defined fibrous capsule along with highly cellular tissue consisting cuboidal or low columnar cells arranged in whorl-like, duct-like, ring-like, ribbon-like or the rosette-like pattern were evident. The duct-like or tubular structure consisted of the central space having eosnophilic material surrounded by a layer of columnar/cuboidal epithelial cells. The nuclei of these cells tend to be polarized away from the central space. Connective tissue stroma revealed areas of hemorrhages with endothelial lined blood vessels and fragments of amorphous calcification [Figure 3].

In the presently reported case, the lesion was removed by surgical enucleation. After reflection of an ample mucosal flap and widened entrance through the usually thinned and expanded cortical bone, the connective tissue capsule of the lesion was encountered. Enucleation was achieved by separation of the lesion from bone without perforating the capsule. Inspection, irrigation, and a gentle curettage of the resultant cavity were done to remove any residual lesion.

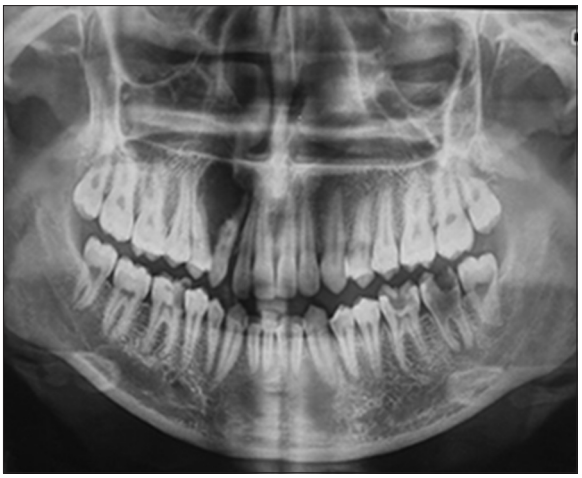

Figure 1: Pear-shaped radiolucency in orthopantomogram

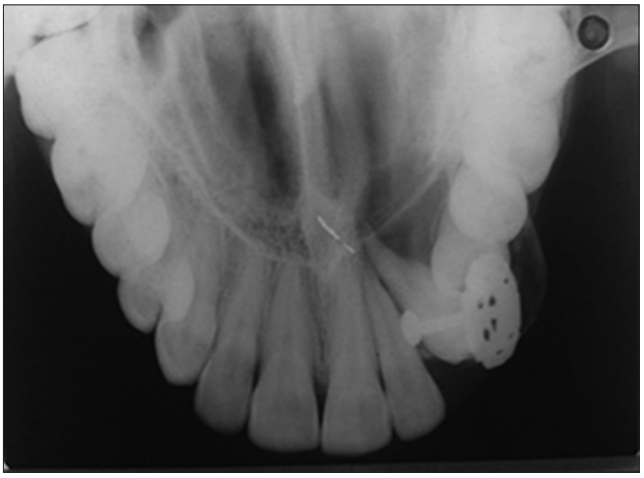

Figure 2: Expansion of buccal cortical plate with root resorption of 23

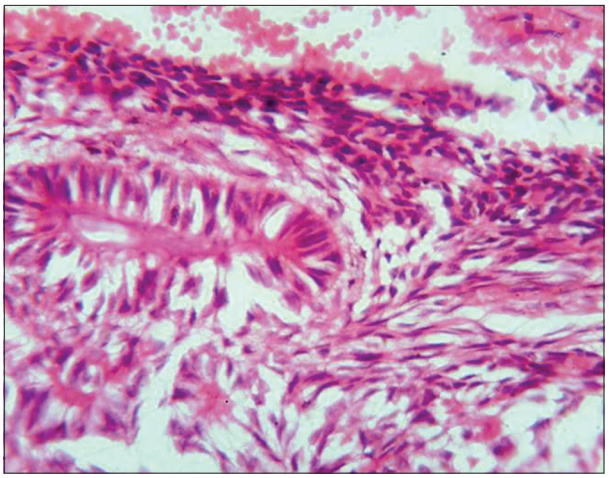

Figure 3: Duct-like pattern with areas of hemorrhage

DISCUSSION

Adenomatoid odontogenic tumor initially described and classified as a variant of the ameloblastoma named as adenoameloblastoma, adenoameloblastic odontoma, pseudoadenomatous ameloblastoma, cystic complex composite odontoma, unusual pleomorphic adenoma-like tumor, ameoloblastic adenomatoid tumor, odonto-ameloblastic tumor, odonto-ameloblastic odontoma, tumor of the enamel organ, ameloblastic epithelial tumor, and tumor connected to development cysts.[6-8] Pindborg named it as AOT, classifying it as an odontogenic epithelial tumor presenting inductive effect at the connective tissue.[9,10]

Adenomatoid odontogenic tumor is a benign, slow growing, epithelial tumor showing an intraoral and extraoral component. Patients are usually present with the impacted teeth within the lesion. The enclosed tooth located within the lesion is usually a canine. The lesion may contain only the crown of the tooth or a whole tooth. It usually occurs in the anterior region of the maxilla, but it may also arise from the mandible.

In this case, the lesion was at the anterior maxilla without any impacted tooth, which is not consistent with the literature. AOT usually has a radiolucent appearance upon radiologic examination, but in some cases, small radiopaque spots (calcifications) resembling snow fiakes may be observed.

In this case, radiologic examination showed pear-shaped radiolucency in between canine and premolar.

WHO has described the histologic features of the tumor as follows: "A tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue.[10] The tumor may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm." The histologic appearance of all variants is identical and exhibits remarkable consistency.[11,12]

At low magnification, the most striking pattern is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue. Between the epithelial cells of the nodules and in the center of the rosette-like configuration eosinophilic amorphous material is often found and described as tumor deposits. Conspicuous within the cellular areas are structures of tubular or duct-like appearance. A third characteristic cellular pattern consists of nodules of polyhedral, eosinophilic epithelial cells with squamous appearance and exhibiting well-defined cell boundaries and prominent intracellular bridges. These islands may contain pools of the amorphous amyloid-like material and globular masses of calcified material (thus, the suggestion of a combination of calcifying epithelial odontogenic tumor and AOT).[13] Another epithelial pattern has a trabecular or cribriform configuration. Occasional foci of mitotic activity can be traced. Induction of hyaline, dysplastic dentinoid material, or calcified osteodentin has been described. Ultrastructurally, tumor epithelial cell types have been recognized, corresponding to the types that are evident on light microscopy.[14] The connective tissue stroma is very loosely structured and contains thin-walled congested vessels characteristically showing marked degenerative (fibrinoid) changes of the endothelial lining, vessel wall, and perivascular connective tissue. It has been suggested recently that the tumor droplets represent some form of enamel matrix.[13]

In the present case, a well-defined fibrous capsule of variable thickness with highly cellular tissue made up of cuboidal or low columnar cells arranged in whorl like, ductular, ring-like and ribbon-like pattern was seen. In the central space of tubular or duct-like pattern, bands of eosinophilic material seen giving a typical rosette pattern appearance. Areas of these cells arranged in a solid pattern were also seen. Connective tissue is scanty with areas of blood vessels and hemorrhages along with areas of amorphous calcification in connective tissue were also seen.

Considerable amount of debate is still going on whether to consider AOT as a hamartoma or neoplasm. The relatively small size of the tumor and lack of recurrences in most cases support the fact that it is a hamartoma. On the contrary, few authors suggest that the early detection could be the reason for small size of the lesion. Therefore, increased variation as compared to the odontogenic apparatus and aggressive features in few of the reported cases certainly gives credibility for the neoplastic origin.[14]

CONCLUSION

Our present case has some unusual features that support its neoplastic nature.

First, AOTs are slow growing and relatively smaller in size, however, some large tumor have been reported. The present case showed unusual recurrence and rapid growth to more than 5 cm within 1 year, as reported by the patient. Second, a well-defined fibrous capsule was present around the tumor. Third, there was considerably root resorption of 23.24, and large areas of hemorrhage were also present.

Thus, based on currently available evidences and clinical and histopathological findings in the present case, we consider AOT as a true neoplasm which is aggressive in nature rather than hamartoma.

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Cite this article as: Bajaj P, Singh G, Garg D, Mohammed RP, Kapoor C, Sabharwal R, *et al.* A true neoplasm: Adenomatoid odontogenic tumor. Clin Cancer Investig J 2015;4:57-60.

Source of Support: Nil, **Conflict of Interest:** None declared.