

Adenomatoid odontogenic tumor, an uncommon tumor

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Abstract

Here we report a case of adenomatoid odontogenic tumor (AOT) in the maxilla in a young girl aged 14 years and its surgical management. We also review the literature and variations in the nomenclature and classifications of this interesting tumor. The review of literature gives an interesting picture regarding terminologies in the past and dilemma in classifying this tumor. The introduction of the name adenomatoid odontogenic tumour has resulted in the simpler and fruitful surgical management like enucleation and curettage with no reports of recurrences. In the past, similar lesion with the terminology like adeno ameloblastoma has resulted in unnecessary mutilating surgery. The conflicting views whether the lesion is being neoplasm or an anomalous hamartomatous growth is also being discussed.

Keywords: Adenomatoid odontogenic cyst, adenomatoid odontogenic tumor, hamartoma

Introduction

Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic lesion that affects young patients associated with an impacted tooth, usually canine. AOT represents 3–7% of all odontogenic tumors.^[1-3]

The histogenesis of AOT is still uncertain and sometimes categorized as a hamartomatous lesion. The tumor is sometimes referred as Two Third's tumor because it occurs in the maxilla in about 2/3 cases, about 2/3 cases in young females, 2/3 case associated with impacted tooth, 2/3 case affected tooth is canine.^[4]

Review of Literature

In the past, various names have been used for cases similar to AOT in the literature.

In 1909 James and Forbes from England reported a case of epithelial odontome which is similar to AOT.^[5]

In 1915 Harbitz of Norway reported a case of cystic adamantoma.^[5]

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In 1916 Wohl of Omaha reported AOT-like case as tooth germ cyst of the jaw (or chorioblastoma) ^[5]

In 1948 Stafne reported first series of AOT under the title epithelial tumors associated with developments cyst of maxilla.

Bernier and Tiecke were the first to publish a case using the name adeno ameloblastoma.^[6]

In 1968 Abrams *et al* suggested the term odontogenic adenomatoid tumor.

In 1969 Philipson and Birn proposed the name adenomatoid odontogenic tumor.^[7]

Later the AOT was adopted in the initial edition of World Health Organization (WHO)'s histological typing of odontogenic tumor, jaw cysts, and allied lesion in 1971^[8] and retained in the 2nd edition of WHO in 1992.

In 1998 Reichart and Philipsen presented the update on AOT based on more than 600 cases in their recent reference work "Odontogenic Tumour and Allied Lesions".^[9]

Case Report

A 14-year-old female child reported to our clinic with a complaint of missing teeth in the right maxilla with a medical history taking epileptic drugs.

Extra oral examination

There is diffused swelling involving right anterior maxilla with moderate obliteration of nasolabial fold [Figure 1].

Intra oral examination

There is a smooth circumscribed swelling 2 cm × 3 cm size, with well-defined margins in the right maxillary region obliterating the buccal vestibule. The buccal cortex is expanded and the surface of the swelling is smooth. Mild

tenderness is present. The consistency is firm and fluctuation of the cortex in one area is present. The teeth 13, 14, and 15 were missing. Aspiration of the swelling yielded 2 ml of straw color fluid mixed with blood.

Radiographic findings

Well-circumscribed radiolucent lesion same in right maxilla with impacted teeth 13 and 14, with well-defined radio-opaque border. [Figure 2] Radiograph clearly showed the juxta position of the impacted canine and premolar with flakes of calcifications inside the lesion [Figure 3].

Computed tomography findings

Well-defined cystic lesions present with impacted premolar and canine. There are flakes of radio-opaque areas inside the lesion

Differential diagnosis

1. Dentigerous cyst
2. Adenomatoid odontogenic tumor
3. Calcifying odontogenic cyst

Discussion

AOT usually occurs within the tooth bearing areas of jaws and often found in association with impacted teeth.

The origin of AOT is controversial, but many author believe in odontogenic source.

AOT has cytological features similar to various components of enamel organ, dental lamina, reduced enamel epithelium, and its remnants.^[4]

Radiographically, AOT frequently looks like a dentigerous cyst. The lesion is usually unilocular and radiolucent. However, they contain fine calcifications (snowflake), a feature that may be helpful in differentiating an AOT from dentigerous cyst. The unilocular radiolucency is well demarcated with smooth cortical border. Most lesions are pericoronal, juxta coronal, and divergence of roots and displacement of teeth often occurs without root resorption.^[2,3,10,11]

Radiographic differential diagnosis:^[4]

1. Dentigerous cyst
2. Calcifying odontogenic cyst
3. Calcifying odontogenic tumor
4. Uni cystic ameloblastoma
5. Odontogenic kerato cyst

Microscopic features:^[2]

AOT is usually surrounded by a well-developed connective tissue capsule. It may present as a solid mass, a single large cystic space, or as numerous small cystic spaces. The tumor is composed of spindle-shaped or polygonal cells forming sheets and whorled masses in a scant connective

tissue stroma. Between the epithelial cells as well as in the center of rosette-like structures is amorphous eosinophilia material. The characteristic duct-like structures is lined by a single row of columnar epithelial cells, the nuclei of which are polarized away from the central lumen. The lumen may be empty or contain amorphous eosinophilic material. Dystrophic calcifications in varying amounts and in different forms are usually encountered in AOTs, within the lumen of



Figure 1: Intra-oral view



Figure 2: OPG showing circumscribed radio-lucent lesion with calcifications and impacted teeth

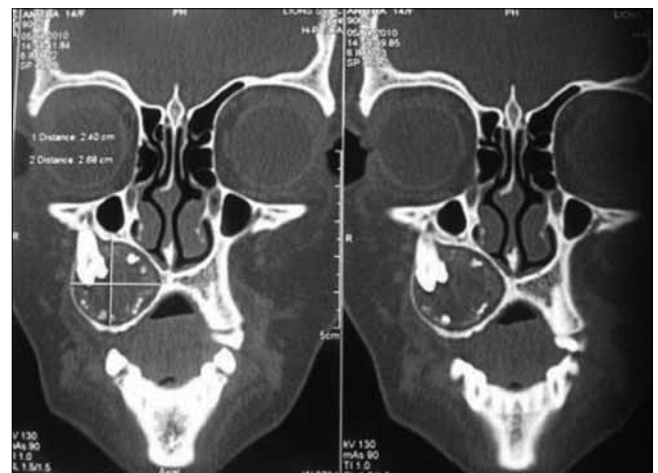


Figure 3: CT—coronal sections

duct-like structures, scattered among epithelial masses, or in the stroma.^[4]

Summary and Conclusion

Even though enucleation and curettage for AOT is the most common treatment modality, accurate histological diagnosis is mandatory to avoid unnecessary mutilating surgery.

Still the search for accurate classification and ideal nomenclature for AOT continues.

The debate as to whether AOT is an anomalous hamartomatous growth or a true benign neoplasm has not been settled yet.^[1] Immunohistochemical studies by certain authors reinforce the theory of hamartomatous character of this lesion indicating AOT is not a true neoplastic lesion.^[12]

Some authors prefer to disagree with the term AOT.^[13] According to Marx and Stern, the more appropriate term is adenomatoid odontogenic cyst (AOC).^[13] The term adenomatoid odontogenic cyst as suggested by Marx and Stern is controversial. But in our case presented, the presence of unilocular cystic lesion, fluid on aspiration, and cystic cavity on transection has to some extent support the terminology adenomatoid odontogenic cyst (AOC) as termed by Marx and Stern.^[13]

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