CASE REPORT

Masquerading swelling of the Maxilla: A case report with review of literature.

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ABSTRACT

Odontogenic cysts and tumors constitute an important aspect of maxillofacial pathology. Although cysts are common tumors by contrast are relatively uncommon. Adenomatoid odontogenic tumor (AOT) represents 3% to 7% of odontogenic tumors. This case report highlights the clinical and histological features and surgical management of adenomatoid odontogenic tumor in the maxilla of a 27-year old patient. The cystic tumor within the maxillary sinus was completely enucleated along with the retained tooth. Healing was uneventful and no local recurrence was observed during the period of follow-up.

INTRODUCTION:

The adenomatoid odontogenic tumor (AOT) was first described by Harbitz¹ in 1915 under the name of cystic adamantoma. Many authors and researchers like Steensland,² Dreybladt,³ and James and Forbes⁴ reported the first case of AOT. In 1971, WHO adopted the term “adenomatoid odontogenic tumor”.⁵ Currently, WHO defines AOT as a tumor with a variety of histological presentation wherein the odontogenic epithelium, embedded in a mature connective tissue stroma and distinguished by its slow but progressive growth.⁶

Adenomatoid odontogenic tumor is a benign odontogenic tumor with a relative frequency of 2.2% to 7.1%.⁶,⁷ This tumor is mostly seen in young patients, in the second decade of life, and is rarely seen in patients older than 30 years of age with female predilection.⁸,⁹,¹⁰ AOT often occurs in the anterior maxillary region, almost twice as often as the mandible and impacted maxillary canine is most commonly associated with AOT.¹¹ Clinically, AOT presents as a slow-growing symptom-free lesion and is frequently discovered during routine radiographic examination.¹²

Occurrence of adenomatoid odontogenic tumor has been described both intraosseously and extraosseously. Over 96% of the cases of this lesion are intrabony and the peripheral variant is rare.¹³ Intraosseous variant has been classified radiographically into 2 types: follicular (or pericoronal) and extrafollicular (or extracoronal) types. Both types presents as well-defined unilocular radiolucency. Follicular type surrounds the crown and is often part of the root of an unerupted tooth whereas the extrafollicular is located either between, above, or superimposed upon the root of an unerupted tooth.

CASE REPORT

A 28 year old female was referred to the Department of Oral and Maxillofacial Surgery of M. S. Ramaiah Dental College and Hospital for evaluation of a swelling in her right cheek region with ipsilateral nasal obstruction. Patient complained of pain and swelling in her right cheek from past one month. She also complained of right preauricular pain radiating to her neck on same side. She gave history of having visited our centre about a year ago with the same complaint but the swelling was not evident extraorally then. Past history revealed history of tooth extraction in the same region due to pain and mobility. Swelling persisted even after a month of extraction, and hence was referred to our centre for opinion. Patient underwent biopsy for the same and it was reported to be fibrous dysplasia. She however did not report to undergo scheduled surgical procedure (due to personal reasons). Presently on examination, single solitary swelling measuring about 2x3cm was observed on the right side of her cheek. It was extending from the right lateral nasal border to anterior border of zygomatic bone laterally and superiorly from infraorbital margin to 1 cm above the right corner of the oral commissure. (Fig. 1) Overlying skin was smooth and
pinchable. Intraoral, diffuse enlargement of the right maxillary edentulous residual ridge extending from distal aspect of right maxillary canine to maxillary tuberosity and 0.5 cm on either sides of the ridge bucco-palatally was observed. Right buccal vestibule corresponding to the premolar and molar region was obliterated. Swelling was ovoid in shape with irregular margins. Surface appeared to be smooth with a normal appearing overlying mucosa with respect to the lesion as well as adjacent mucosa. Indentations of the lower posterior teeth were noticed over the crest of the ridge. No ulceration or discharge was noticed. (Fig. 1)

Computed tomography (CT) scan demonstrated a large radiolucent lesion in relation to right maxilla, with displacement of left maxillary sinus medially and superiorly and thinning of the bony sinus floor. (Fig. 3) An unerupted tooth was seen within the lesion just below the floor of orbit (Fig. 2). There was no contributory medical history and no lymph node involvement was detected. The results of all hematological studies were all within normal limits. Diagnostic biopsy was performed from the left upper vestibular region corresponding to the premolar region and was reported as adenomatoid odontogenic tumour.

After a thorough pre anesthetic work up and informed consent for surgery, patient was posted for en block resection procedure under general anesthesis. Under strict aseptic precautions intra oral gingival crevicular incision was given from right lateral incisor and canine and crestal incision over the swelling till the third molar and a releasing incision was given anteriorly and a complete mucoperiosteal flap was raised both buccally and palatally. The lesion was exposed adequately on the buccal side. Based on the CT scan, the lesion was extending from the periapical region of the right canine and hence ostetomy cut was placed from the socket of lateral incisor after its extraction, extending along the margin of the lesion supero posteriorly along the anterior wall of maxillary sinus, posteriorly below the zygomatic buttress till the third molar region. (Fig. 4, 5) Palatally there was cortical thinning present and hence osteotomy line extended from the extraction socket of lateral incisor 2mm away palatal from the margin of the lesion. The specimen was removed in toto from the maxilla along with the tooth on its superior border. Primary closure was achieved. The specimen submitted for pathological examination consisted of a lesion measuring 6x5x5 cm. (Fig. 6) The postoperative course was uneventful. An immediate obturator was given to protect the surgical site. Later tooth bearing prosthesis was given and planned for cast partial denture. (Fig 9, 10)
Histologic examination on sectioning revealed the characteristics of AOT. The tumour mass was encapsulated by a thick connective tissue capsule. Various-sized solid nodules of cuboidal or columnar epithelial cells formed nests or rosette like structures. Conspicuous within the cellular areas were structures of tubular or duct-like appearance (Fig. 7, 8). Between the epithelial cells and the rosette-like configurations, eosinophilic fibrillar amorphous material and/or calcified masses of various shapes were seen.

**DISCUSSION**

Adenomatoid odontogenic tumor was first recognized as a distinct entity by Stafnes in 1948. In 1991, Philipsen et al comprehensively reviewed the world literature on AOT and found that 45.5% of all documented cases were reported in Asian countries. The adenomatoid odontogenic tumor (AOT) is an infrequent benign epithelial tumor, preferentially found in children and young adults. Both intraosseous and extra osseous forms are distinguished. The subtyping of AOT is based on clinical and radiological findings. The follicular (intraosseous) type is by far the most frequent type of AOT. Adenomatoid odontogenic tumours, account for about 3% of all odontogenic tumors, than odontoma, cementoma, myxoma and ameloblastoma. It was suggested that this tumour may be a hamartoma rather than a true neoplasm, however there is currently no evidence available.

Any corticated radiolucencies with small radiopaque foci, especially in teenagers and young adults, adenomatoid odontogenic tumour should be considered as a differential diagnosis. It is a slowly growing lesion, with a predilection for the anterior maxilla of young females. 69% of these tumors are diagnosed in the second decade of life, and more than half occur during teenage. The female to male ratio for AOT in all age groups is 2:1. This tumor is also referred to as “two third tumor” since 2/3 occur in maxilla, 2/3 in young females, 2/3 associated with unerupted tooth and 2/3 affected teeth are canines.

In the presented case it was a female patient in her 3rd decade of life with the lesion presenting in the right maxillary sinus associated with an impacted second premolar. These tumours enlarge to a size of 1–3 cms in its greatest diameter, but they can be larger, as in our case reported here the size noted was 6x5x5 cm involving the entire maxillary sinus. These lesions are typically asymptomatic, but central lesions results in cortical expansion, as seen in our present case. The involved teeth are commonly impacted, and adjacent teeth may be displaced. The distribution of unerupted teeth associated with the follicular type has a typical pattern. The canines account for 59% of cases and the maxillary canines alone for 40%. However in our case premolar was impacted. Unerupted first and second molars and deciduous teeth are rarely involved. The origin of adenomatoid odontogenic tumours is controversial however some authors believe that they originate from the odontogenic epithelium of a dentigerous cyst.

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. 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.”

This present report on the case of an AOT demonstrated cuboidal to tall columnar odontogenic epithelial cells in mature connective tissue stroma. The odontogenic epithelium was arranged predominantly in the form of solid nests, also thin interconnecting strands and duct like structures containing eosinophilic fibrillar material were found. Focal areas of microcyst formation with eosinophilic tumor droplets and pale eosinophilic fibrillar material were seen.
Initial biopsy could have been mistakenly diagnosed as fibrous dysplasia with a clinicopathological co-association, as the pathology department reported to have received overlying bone from the lesion for histopathological examination. Later the second biopsy which was done a year later included both hard as well as soft tissue from the lesion which confirmed the lesion to be AOT.

Since all variants of AOT presents benign biological behaviour and almost all are encapsulated, conservative surgical excision or curettage is the treatment of choice. However recurrence have been reported in few cases. If the follicle is found to be uninvolved, it can be easily separated from the tumors; it may then be possible to remove the lesion while leaving the teeth in place, as described by Toida and others. However, in the case reported here, the tooth had been pushed almost to the floor of orbit of ipsilateral side, and the large size of the lesion justified the decision to remove the tooth along with the lesion.

**CONCLUSION**

Adenomatoid odontogenic tumor is a benign epithelial odontogenic tumor that is frequently seen in the anterior maxillary region. It is more frequently associated with an impacted tooth. Hence it is most commonly diagnosed as dentigerous cyst. It is a slow growing, painless swelling and often diagnosed coincidently. Since most of these variants of AOT are benign and innocuous conservative surgical excision or curettage is the treatment of choice with very low rate of recurrence.

**REFERENCES**