

Adenomatoid Odontogenic Tumor –An unusual Case report in a 40 years old female patient

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Abstract

Adenomatoid Odontogenic tumor is a benign epithelial and slowly growing tumor which accounts for about 3% to 7% of all the Odontogenic tumors of oral cavity. Most commonly encounter during 2nd and 3rd decade of life with female predisposition. Commonly found in anterior maxilla with impacted canine. The present case is a rare case because the AOT encounter was of anterior mandible which is not usually found with the incidence of the AOT. AOT is a true benign neoplasm which causes facial deformity with a very less recurrence rate of 0.2%. This tumor is also called as “two thirds tumor” because it occurs in in maxilla in 2/3 cases, around 2/3 cases in young females, 2/3 cases associated with un erupted teeth

and 2/3 affected teeth are canines. It is classified in to follicular and extra-follicular varieties. Our case is rare case of extra-follicular variety which shows resorption of multiple teeth roots and author recommends removal of this tumour in TOTO.

Keywords: Impacted teeth, AOT, true neoplasm, facial deformity

Introduction

The adenomatoid Odontogenic tumor is a rare tumor that comprises only 0.1 per cent of tumours and cysts of the jaw and 3% of all Odontogenic tumors. The AOT is a slowly growing benign hamartomatous lesion which is not tending to spread widely. Mostly occurs in intraosseous

and in peripheral forms, with follicular and an extra follicular types

AOT is common in anterior maxilla than posterior region with female predilection & commonly encounters during second and third decade of life .It shows a prevalence of about 1.2% in Caucasian and 9% in black African patients The AOT is over two times more commonly located in the anterior maxilla than the mandible. Normally the upper canine and when in mandible shows lower canine region predilection. Philipsen and Brin coined the term AOT, which was later adopted by the WHO classification in 1971.^(1,2) letter on various synonyms were used for same entity as Adenoameloblastoma Adenomatoid ameloblastoma, Adenomatoid odontogenic tumor, Pleomorphic adenomatoid tumor⁽³⁾ The basic moto for presenting this case was to document the clinical presentation and treatment protocols for AOT of jaws, which may become an addition in literature.

Case report

A 40 years old Female patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of symptomatic swelling over chin region of the face since last 2 months. On general examination, patient was well oriented to time, place and person, with mesomorphic built, Normal gait, well nourished, with weight of 58 kg. No signs of anaemia, icterus, cyanosis, clubbing and pedal oedema were present. Right submandibular lymph nodes were palpable and tender on palpation. No significant past medical, Familial and dental history was contributory. There was no history of trauma to the site of chief complaint; no history of pus discharge was reported by the patient.

On clinical examination Extra-oral findings revealed well defined swelling over symphysis of mandible extending bilaterally towards parasymphysis with slight mandibular prognathism, which was tender, non-reducible, non-

compressive in nature while cracking sound was also noted and swelling was soft in consistency with raised localized temperature. Parasthesia was noted on left Para symphysis region.

While intra-oral examination showed buccal cortical expansion without lingual cortical expansion and extending antero-posteriorly from lower right 2nd premolar to left first molar tooth.(**fig 3**)The overlying mucosa was normal, on palpation the region was mild tender with mobility of 1st lower right premolar till lower left 1st molar.

Initially Orthopentomograph was taken which showed unilocular radiolucency extending from right 2nd premolar till left 1st molar extending till lower border of mandible with impacted and pathologically migrated tooth with resorption of all involved teeth was noted with thinning of lower border of anterior mandible. Hence Cone Beam Computed Tomography scan was taken which was showing hypo dense unilocular region involving anterior mandible. Large hypo-dense unilocular area was extending from right side of mandible till left side with an impacted left canine with pathological migration toward lower border of mandible was noted and root resorption of all the involved teeth was noted suggestive of Odontogenic tumor. There was no evidence of lingual plate expansion. This lesion measured was approximately of 33.1 x 12.9 mm. (**Fig 2**) by analysing these clinico-radiological findings, the lesion was interpreted as adenomatoid Odontogenic tumor.

Initially aspiration of the cystic region was done which came out positive and showed straw color fluid followed by Incisional biopsy of the large cystic lesion of the mandible under local anesthesia was taken which revealed adenomatoid Odontogenic tumor. On histopathological diagnosis, which was surgically enucleated in Toto along with removal of impacted canine and extraction of all the

involved teeth successfully under local anesthesia. The enucleated cystic linings were again sent for Histopathological examination which confirmed the diagnosis of adenomatoid Odontogenic tumor with sheets of polygonal cells throughout the fibrous connective tissue stroma with amorphous calcified material was present. postoperative course was uneventful. Patient was kept on normal antibiotics, NSIDS and antacids for 5 days. Letter on rehabilitation with removable partial denture was done post-operatively after 1 month. Patient was kept under follow up for 3 years and no recurrence is found till date. With respect to the age of the patient and the localization of the AOT in the lower jaw, the reported case is a rare example of this tumor entity.

Discussion

The adenomatoid odontogenic tumour was first recognized by Stafne in 1948.⁽⁴⁾ In 1992 WHO defined it as: "A tumor of odontogenic epithelium with duct like structures and with varying degrees of inductive change in the connective tissue". It has high female predilection in the age range of 20–30 years.^(5,6) Adenomatoid odontogenic tumour is a slowly growing lesion, with a predilection for the anterior maxilla (ratio of cases 2:1 relative to mandible) of young females. 69 % of adenomatoid odontogenic tumours are diagnosed in the second decade of life, and more than 50% occur during the teenage years. The female to male ratio for all age groups and all variants is close to 2:1.⁽⁷⁾ AOT usually occurs within the tooth bearing areas of jaws and often found in association with impacted teeth. In few exceptional cases the tumour is associated with unerupted teeth. The unerupted teeth are usually canine or lateral incisors.⁽⁴⁾ The origin of AOT is controversial, but many authors believe in odontogenic source. AOT has cytological features similar to various components of enamel organ, dental lamina, reduced enamel epithelium, and its

remnants.⁽⁸⁾ Philipsen and Brin coined the term AOT, which was later adopted by the WHO classification in 1971.⁽¹⁾ But some authors prefer to disagree with the term AOT.⁽⁹⁾ The term adenomatoid odontogenic cyst as suggested by Marx and Stern is controversial. But in our case, the presence of unilocular cystic lesion, fluid on aspiration, and cystic cavity on transection support the terminology as an adenomatoid odontogenic cyst. 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.⁽¹⁰⁾ Irregular root resorption and dilaceration within the lesion are only infrequently reported in the literature. Clinical, radiographic and macroscopic findings in present case are consistent with descriptions of the lesion in the dental literature. In past studies after enucleation of the tumor with follow up of 5 to 10 years showed no evidence of recurrence.⁽⁴⁾ **Giansanti et al** reported that the adenomatoid Odontogenic tumor was a completely benign tumor which never recurred once removed. This tumor is also called as "two thirds tumour" because it occurs in in maxilla in 2/3 cases, around 2/3 cases in young females, 2/3 cases associated with un erupted teeth and 2/3 affected teeth are canines. It is classified in to follicular and extra-follicular varieties.⁽¹¹⁾ Our case was of extra-follicular type which showed resorption of all the involved teeth. Presented case should be considered to be rare case because of the age which is 40 years in our case while normally it occurs in between 2nd and 3rd decade of life, and the lesion

was of large size extending from right side of mandible till left side and it was found in mandible than in maxilla.

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⁽¹²⁾. The debate as to whether AOT is an anomalous hamartomatous growth or a true benign neoplasm has not been settled yet. Immunohistochemical studies by certain authors reinforce the theory of hamartomatous character of this lesion indicating AOT is not a true neoplastic lesion.⁽¹³⁾ We should always consider the lesion as a benign tumor because its locally aggressive causes resorption of involved teeth and facial disfigurement along with causes involved nerve displacement and nerve involvement can leads to parasthesia which is a sign of a true tumor. Along with it ,causes bony resorption hence early diagnosis and complete enucleation with preservation of involved important anatomical landmarks is mandatory.

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Legend Figure



Fig 1: Extra-oral findings revealed well defined swelling over symphysis of mandible extending bilaterally towards parasymphysis



Fig 2: Orthopantomograph was taken which showed unilocular radiolucency extending from right 2nd premolar till left 1st molar extending till lower border of mandible with impacted and pathologically migrated tooth with resorption of all involved teeth was noted with thinning of lower border of anterior mandible

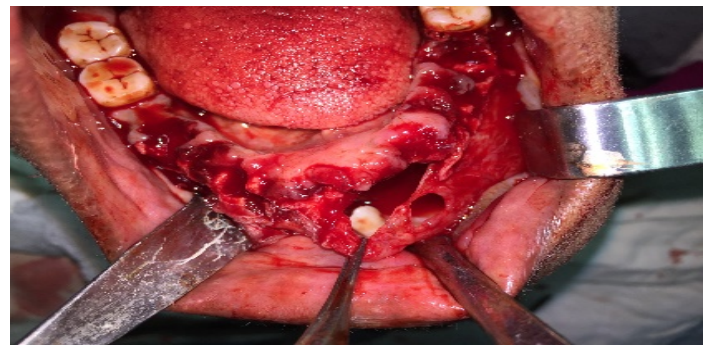


Fig 3: Intra- operative view showing impacted canine



Fig 4 : Post Operative 3 year follow up