Case Report

Indian Journal of Dental Education
Volume 8 Number 4, October - December 2015
https://dx.doi.org/10.21088/ijde.0974.6099.8415.8

Adenamatoid Odontogenic Tumour

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Abstract

Adenomatoid odontogenic tumor was first described by Steensland in 1905. It is an uncommon benign tumor of odontogenic origin with an incidence of 2.2 to 7.1%. The lesions are typically asymptomatic but may cause cortical expansion and displacement of the adjacent teeth. AOT can occur both intraosseously and extraosseously. This case report describes intra-osseous counterpart of adenomatoid odontogenic tumor.

Keywords: Odontogenic Tumors; Adenomatoid Odontogenic Tumors; Intra-osseous; Asymptomatic; Panoramic Radiograph.

Case Report

A 14 year old girl (Fig.1) reported to Department of Oral medicine and & Radiology with presenting complaint of swelling involving right side of face since 6 months. The swelling was progressively increasing in size and causing facial asymmetry. There was non-relevant past medical history and dental history. There was no significant family history recorded. She had normal gait, posture, well oriented to surroundings and normal intelligence. There was no history of pallor, clubbing, cyanosis and lymphadenopathy noted. The vital signs were within normal limit and having no history of any systemic diseases. On extra-oral examination (Fig.2) a diffuse swelling of 4x4 cm noted involving right maxillary area. Superiorly it was extending from infra-orbital margin to upper lip inferiorly causing drooping of right half of upper lip. Medially swelling was extending along right lateral nasal wall to ala of nose causing complete obliteration of nasolabial fold and displacement of right ala of nose. Laterally swelling was extending till line along outer canthus of eye involving right maxillary area. On intra-oral examination (Fig. 3) a hard, non tender swelling of 4x3 cm extending from right maxillary central incisor to right maxillary 2nd premolar area noted. The swelling was causing massive buccal cortical expansion without any lingual cortical plate involvement leading to obliteration of right mucobuccal fold. There was no sign of fluctuancy, compressibility or reducibility. There was no associated bleeding, discharge, pain or local rise of temperature. The overlying mucosa was smooth and had normal appearance. The patient was advised for panoramic radiograph. The panoramic radiograph (Fig. 4) shows a well-defined radiolucency extending from the right maxillary central incisor to the right maxillary 1st premolar with a retained deciduous lateral incisor and deciduous canine. The permanent maxillary right lateral incisor and canine are impacted. The maxillary canine is displaced lying apical to the roots of the right maxillary permolar and the lateral incisor lies in the lateral nasal wall. There is displacement of maxillary right premolars. On the basis of panoramic radiograph findings contrast enhanced CT is advised. The contrast enhanced CT shows a well-defined cystic lesion measuring 3.5x3 cm involving the right maxilla anteriorly. The lesion is extending up to the anterior and medial wall of the right maxillary sinus and right nasal cavity. There are many impacted teeth. The low-density, cystic lesion is surrounded by a thick soft tissue capsule with peripheral enhancement. The lesion is expansile and radiolucent with multiple radiopaque flecks towards the periphery. Small foci of bony dehiscence were noted. Aspiration was uneventful. On the basis of clinic-radiological finding a provisional diagnosis of adenomatoid odontogenic tumor has been made. The differential diagnosis

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includes ameloblastoma, ameloblastic fibroma, calcifying odontogenic tumor or cysts, and ameloblastic fibro-odontoma. The patient is further advised for incisional biopsy and routine blood investigation is carried out. The blood picture was normal. The incisional biopsy is carried out under local anaesthesia. The histopathology revealed an epithelial neoplasm composed of sheets and nests of polyhedral epithelial cells with an abundant eosinophilic , granular cytoplasm. Cellular outlines were distinct and intercellular bridges were noted. Considerable nuclear polymorphism was a frequent finding. Extracellular amyloid-like substance and calcified concentric deposits known as Liesegang rings are also noted (Fig. 9, Fig. 10). On the basis of histopathology final diagnosis of adenomatoid odontogenic tumor has been achieved. The patient is further treated with segmental resection of maxilla and reconstruction. The patient was followed for 1 year but no recurrence has been reported.

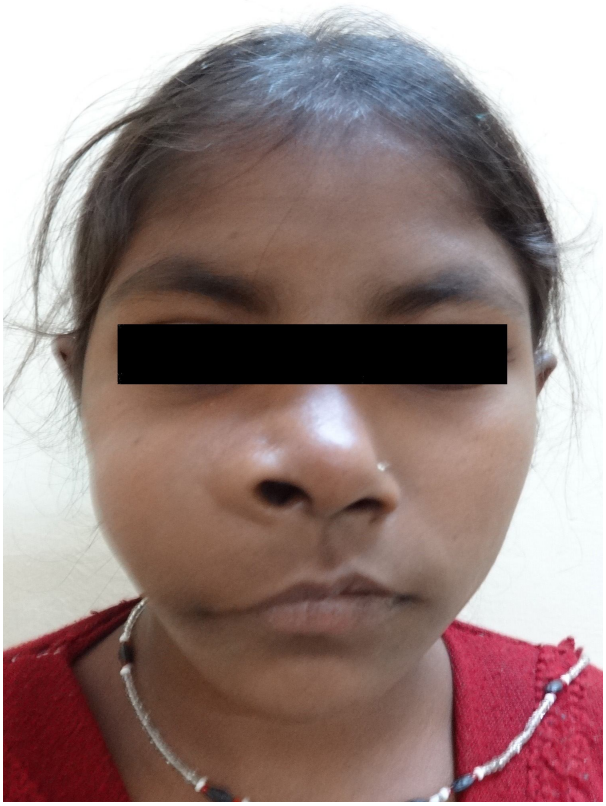

Fig. 1: Profile view of patient

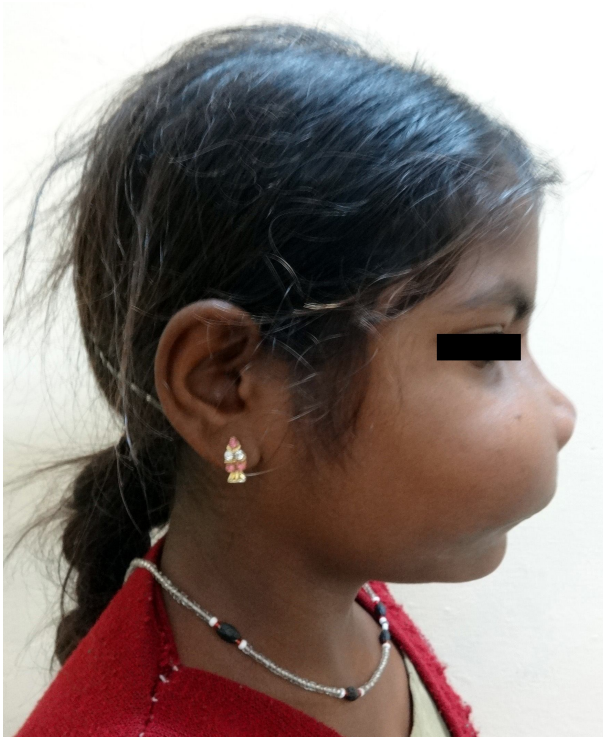

Fig. 2: Extra-oral view of patient showing a diffuse swelling of 4x4 cm involving right maxillary area. Superiorly it was extending from infra-orbital margin to upper lip inferiorly causing drooping of right half of upper lip

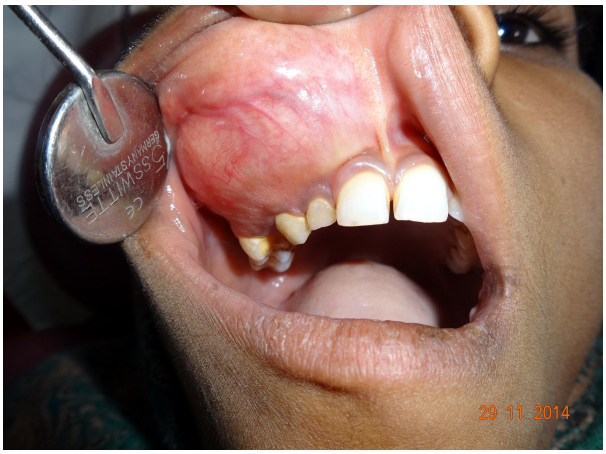

Fig. 3: Intra-oral view showing a swelling of 4x3 cm extending from right maxillary central incisor to right maxillary 2nd premolar area. The swelling was causing massive buccal cortical expansion leading to obliteration of right mucobuccal fold

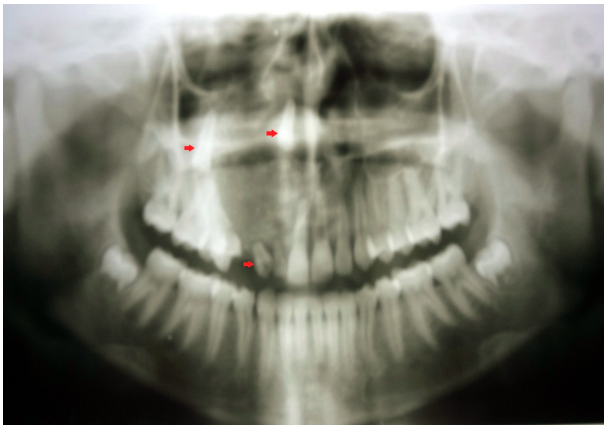

Fig. 4: Panoramic radiograph showing a well-defined radiolucency extending from the right maxillary central incisor to the right maxillary 1st premolar with a retained deciduous lateral incisor and deciduous canine

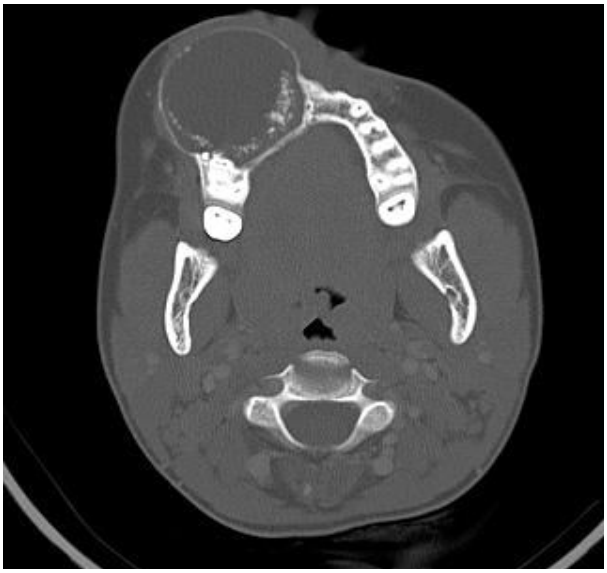

Fig. 5: Axial CT shows a well-defined cystic lesion measuring 3.5x3 cm involving the right maxilla with multiple specks of calcifications

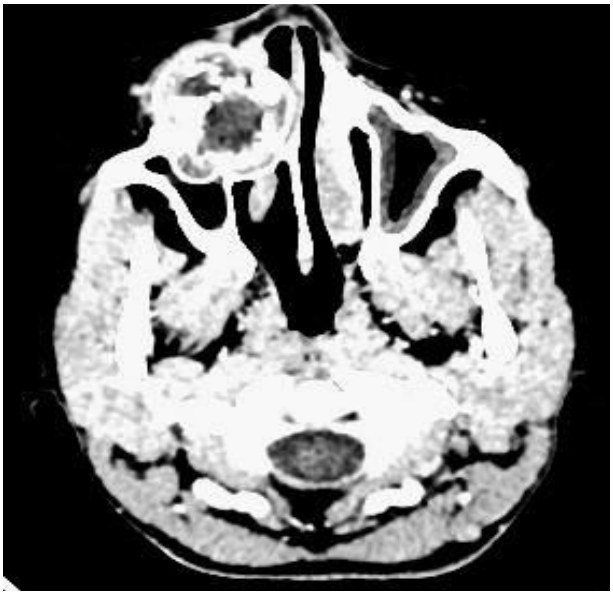

Fig. 6: Axial CECT image showing a well-defined lesion with peripheral enhancement. The lesion is extending up to the anterior and medial wall of the right maxillary sinus and right nasal cavity

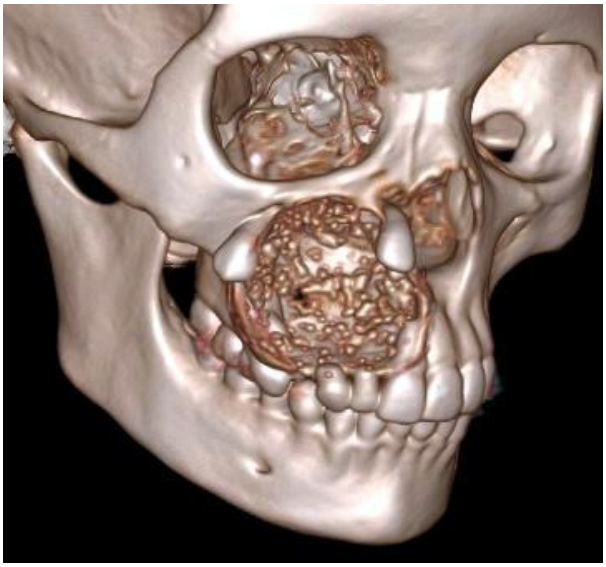

Fig. 7: 3D CT showing displaced permanent maxillary right canine and maxillary lateral incisor with nasal wall involvement and calcifications specks

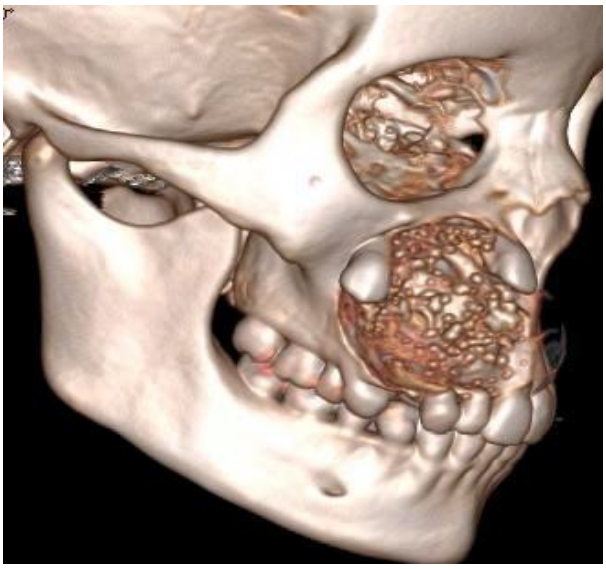

Fig. 8: 3D CT image shows anterior wall involvement of the right maxilla by the adenomatoid odontogenic tumour

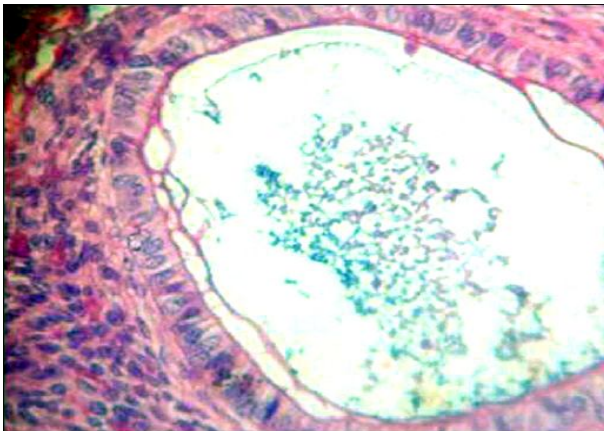

Fig. 9: Hematoxyline and Eosin stained tissue (100x) showing whorls and sheets of cuboidal epithelial cells present in a rosette like arrangements

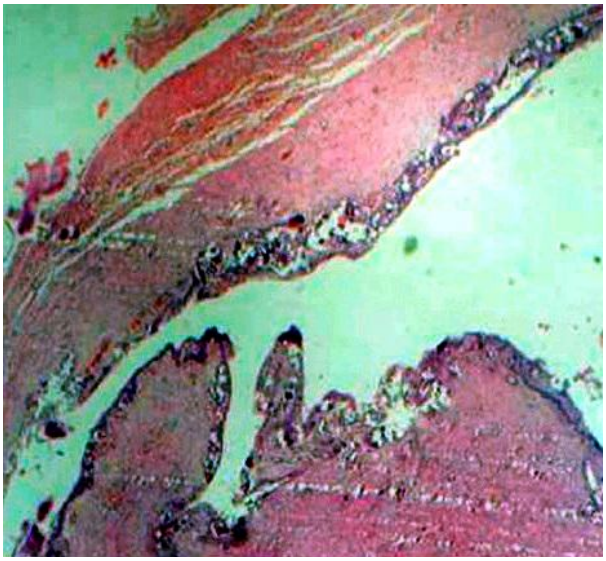

Fig. 10: Hematoxylin and Eosin stained (400x) stained tissue exhibiting a cyst like epithelium made up of cuboidal epithelial cells showing encapsulations

Discussion

Adenomatoid odontogenic tumor (AOT) is an uncommon benign odontogenic lesion that affects young and adolescents. It is usually found in association with an impacted tooth and most commonly associated with impacted canine. Adenomatoid odontogenic tumor represents 3–7% of all odontogenic tumors [1].This tumor is also referred as Two Third's tumor become 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 [2]. Adenomatoid odontogenic was first documented in literature by Steensland as epithelioma adamantinum [3].The WHO histological typing of odontogenic tumors, jaw cyst and allied lesions (2005) has defined adenomatoid odontogenic tumor as a tumor of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue [4]. In 1995 Unal et al. produced a list containing all nomenclatures for adenomatoid odontogenic tumor reported in the literature like adenoameloblastoma, ameloblasticadenomatoid tumor, adamantinoma, epitheliomaadamantinum, or teratomatousodontoma [5]. AOT has more predilection for females than male in ratio of 2:1 [6]. They have peak incidence in the second decade of life and uncommon in patients above 30 years of age [7]. In 1969, Philipsen and Brin proposed the name AOT which was widely accepted and this term was adopted by first edition of the World Health Organization (WHO) in 1971 [8].There are three variants of adenomatoid odontogenic tumor, follicular variant (73%), which has a central lesion associated with an embedded tooth, extrafollicular variant (24%) which has a central lesion and no connection with the tooth and the peripheral variety (3%) [9].

Clinically, Adenomatoid odontogenic tumor is a slow growing, asymptomatic swelling associated with unerupted tooth. It is usually discovered during routine radiographic examination [10]. However delayed eruption of tooth or slow growing bony expansion with or without displacement of adjacent teeth commonly leads to discovery of adenomatoid odontogenic tumors during routine radiography. AOT can cause tooth mobility, jaw bone expansion and asymmetrical facial swelling [11]. Displacement of teeth due to tumor expansion is much more common than root resorptions. The peripheral lesions may show some erosions of the adjacent cortical bone [12]. Radiographically, AOT usually appears as unilocular lesion which contain fine radiopaque calcifications and irregular root resorption is rare [13]. The radiopaque calcifications are described as flecks, snow-flakes, and patchy areas of calcification, scattered radio-opacities, irregular radioopacities, amorphous radioopacities, fine radioopacities and faint radio-opacitie [14]. Radiographically, the intraosseous adenomatoid odontogenic tumor usually appears as a pericoronal well circumscribed unilocular radiolucency or radiopaque radiolucent mixed lesion with well defined corticated or sclerotic border usually surrounding an unerupted tooth and may contain multiple minute variable shaped calcifications or radiopaque foci which may appear like a "cluster of small pebbles". These calcified deposits are seen in approximately 78% of the lesions [15]. Differental diagnosis can be ameloblastoma, ameloblastic fibroma and ameloblastic fibro-odontomacalcifying odontogenic tumor or cysts [16].The origin of adenomatoid odontogenic tumor is believed to be from an odontogenic source, the cytological features are similar to those of the enamel organ, reduced enamel epithelium, dental lamina and their remanants [1]. Histologically, the tumor is solid and there is a cyst formation. The epithelium is in the form of whorled masses of spindle cells as well as sheets and plexiform strands. Rings of columnar cells give rise to duct-like appearance. Sometimes extensive calcification is seen [17]. However WHO has described the histologic features of AOT 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 or solid lesion. The histologic appearance of all variants is identical and exhibits remarkable consistency [18]. Conservative surgical enucleation or curettage is the treatment modality of choice.

Guided tissue regeneration with membrane technique is suggested for periodontal intrabony defects caused by AOT after complete removal of the tumor [19]. Malignant potential has not been reported to date. Recurrence of AOT is exceptionally rare [20].

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