



Cone beam computed tomography findings of a large Adenomatoid Odontogenic Tumour in the anterior mandible- A case report

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Abstract

Adenomatoid Odontogenic Tumour (AOT) accounts for 0.1% of all jaw tumours. It commonly occurs in anterior maxilla associated with an impacted tooth. The present case report describes the clinical and radiographic findings of Adenomatoid Odontogenic Tumour occurring in an unusual location. The large size of tumour in short duration, location in the anterior mandible, radiographic absence of calcifications as well as presence of root resorption makes this report distinctive. In addition, the cone beam computed tomography findings of AOT are discussed in detail.

Keywords: Adenomatoid Odontogenic Tumour; Cone Beam Computed Tomography; Mandible.

INTRODUCTION

Adenomatoid Odontogenic Tumor (AOT) is a benign lesion of odontogenic origin. The first ever reported case of AOT was in 1905 by Steensland as "epithelioma adamantinum". (1) AOT is considered as a hamartoma rather than a neoplasm (2) having slow progressive growth and accounting for 2.2–13% of all odontogenic tumors (3). Hamartomas can be defined as a non-neoplastic, unifocal or multifocal developmental malformation, comprising a mixture of cytologically normal mature cells and tissues which are indigenous to the anatomic location, showing disorganized architectural pattern with predominance of one of its components (4). Adenomatoid Odontogenic Tumour is also referred to as "tumor of two-third" because two-third of these cases occur in young females, two-third in the maxilla, two-third are associated with unerupted teeth, two-thirds are associated with canines and two-

third of the tumors are diagnosed in the second decade of life(2). We describe a case of large extrafollicular type of AOT occurring in the anterior mandible which was of short duration and without radiographic evidence of calcifications.

CASE REPORT

A 39-years-old male reported to the department of oral medicine and radiology with the chief complaint of swelling in the lower jaw since 3 months. The patient gave history of swelling in the left lower third of face which initially presented as a very small swelling and gradually progressed to the present size over a period of 3 months. There was no history of pain and pus discharge or any history of paresthesia. The patient was a known case of tuberculosis and was under treatment during his visit to the hospital. Extraoral examination showed mild facial asymmetry in the lower third of the face with swelling measuring approximately 4 X 5 cms in size and extending from the midline to 0.5 cm away from the left angle of the mouth. Superiorly, the swelling was in line with the left angle of the mouth and inferiorly extended till the lower border of the mandible. No changes were noticed on the overlying skin. On palpation, the swelling was nontender without increase in surface temperature (Figure 1A).

Intraoral examination revealed a bony hard swelling measuring approximately 3.5 X 4.5 cms in size extending from the distal aspect of lower right central incisor till the distal aspect of lower left second premolar.

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Superiorly the swelling extended till the gingival margin of the involved teeth and inferiorly till the lower labial sulcus. Expansion of the buccal and lingual cortex was noted. Overlying mucosa showed no changes (Figure 1B).



Figure 1. A) Clinical photograph showing diffuse swelling on the lower third of the face. B) Intraoral photograph showing bicortical expansion with normal overlying mucosa.

On palpation the swelling was bony hard in consistency with fluctuant areas in the anterior aspect. On palpation there was no pain or paresthesia. Aspiration yielded straw coloured fluid. On electric pulp vitality test, the lower right and left central incisors, lower right and left canine, lower right lateral incisor, lower left second premolar showed no response. The lower left first molar was vital and showed a response at 2. Intraoral examination also revealed grade I mobility with respect to lower left central incisor, lower right central incisor, lower right lateral incisor and grade II mobility with respect to lower left canine, lower left first premolar and lower left second premolar. Missing lower left lateral incisor and lower right second premolar along with restored upper right first molar and decayed lower right first molar were also seen. The Mandibular occlusal radiograph showed bicortical expansion (Figure 2).



Figure 2. Occlusal radiograph showing unilocular radiolucency causing bicortical expansion and displacement of teeth.

Panoramic radiograph showed a unilocular radiolucency measuring approximately 6.2 X 3.1 cms extending from the mesial aspect of lower left first premolar to the distal aspect of lower right second premolar. Displacement of lower left canine and premolars were noted with the

extension of radiolucency till the alveolar crest between the lower left canine and first premolar with intact lower border of the mandible. Lower right central incisor, lateral incisor and canine showed root resorption at the apical third (Figure 3).



Figure 3: Panoramic radiograph demonstrating unilocular radiolucency from mesial aspect of lower left first premolar to distal aspect of lower right second premolar, displacement of lower left canine and premolars and root resorption in mandibular right central incisor, lateral incisor and canine. Measurements are given in mm.

Cone Beam Computed Tomography was done. Coronal section showed unilocular radiolucency in the lower anterior region crossing the midline and extending from the lower right canine till the lower left first molar. On CBCT examination the lesion extended to the mental foramen region on the left side and nerve involvement was not noticed. Axial sections showed expansion, thinning and perforation of both the labial and lingual cortical plates. Expansion and thinning of the inferior border of the mandible was also noted. Root resorption was seen in the lower right central, lateral incisor and canine. No radiopacities were visible within the radiolucency (Figure 4&5).

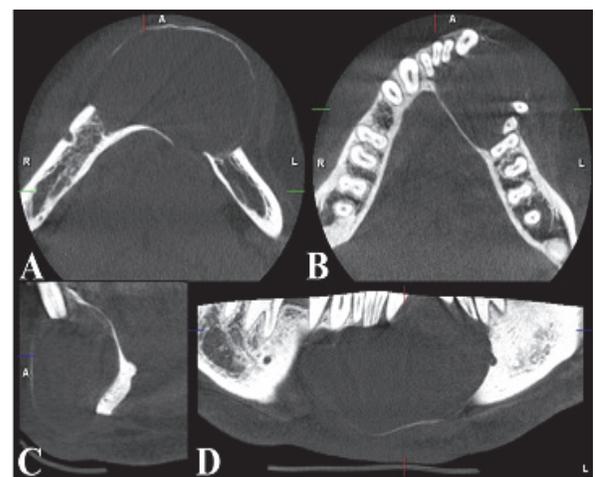


Figure 4. Cone Beam Computed Tomography A) Axial Section showing buccal and lingual cortical perforation. B) Axial section showing unilocular radiolucency with teeth displacement. C) Sagittal Section showing thinning of the inferior border of the mandible with root resorption. D) Panoramic reconstruction showing the extent of the lesion.

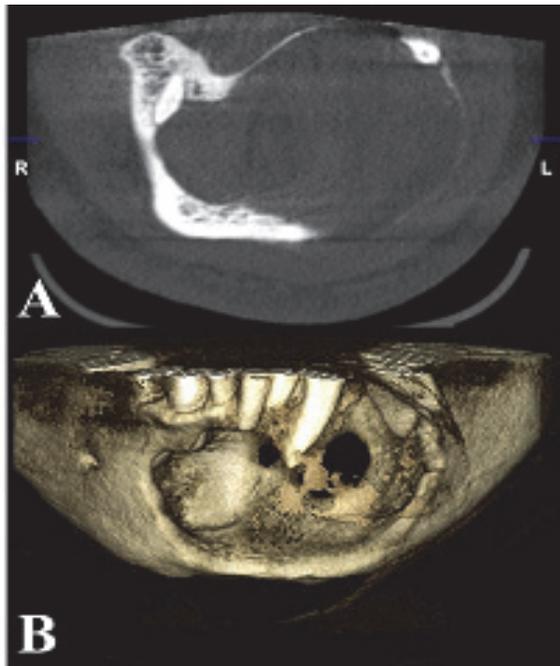


Figure 5. A) Coronal section showing unilocular radiolucency with no calcifications. B) 3D reconstruction showing the extent of the lesion and buccal and palatal perforation.

Based on the clinical and radiographic features, a provisional diagnosis of unicystic ameloblastoma was given. The lesion was surgically excised. Histopathological examination revealed epithelial lining composed of proliferation of odontogenic epithelial cells, that are present in various patterns comprising of duct-like structures, sheets and islands. Rosette like and duct like structures were seen. The duct-like structures showed a layer of eosinophilic hyaline ring (Figure 6). Histopathological diagnosis was given as Adenomatoid odontogenic tumour. Postoperative panoramic radiograph taken after one month showed complete removal of the lesion.

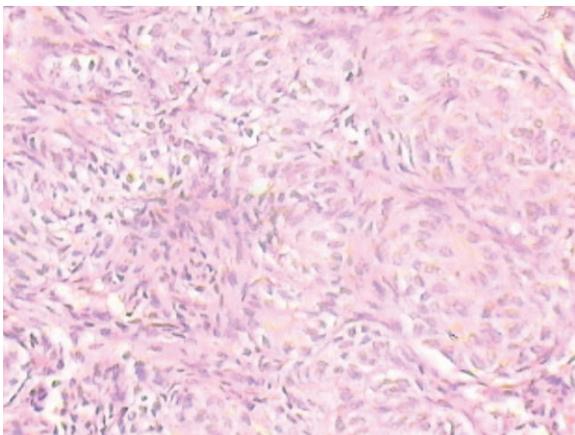


Figure 6. Histopathology (40X) showing odontogenic epithelial cells that are present as duct like structures, sheets and islands.

DISCUSSION

AOT was once known by the following synonyms including Adenoameloblastoma, ameloblastic adenomatoid tumor, teratomatous odontoma and epithelioma adamantinum. In 1948 Philipsen and Birn suggested the term "Adenomatoid odontogenic tumor". In 1971 World Health Organisation accepted this name and defined AOT as "a tumour composed of odontogenic epithelium in various histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow but progressive growth." (5). AOT accounts for 0.1% of all jaw tumours (6). Ratio of incidence of AOT in maxilla to mandible is 1.7:1 (5). The most common site of incidence is the anterior region of maxilla. The female to male ratio for all age groups and all variants is close to 2:1 (6). Our case presented in the anterior mandible in a male subject.

The reported age range is from 3 to 82 years. Greater than 70% of the lesions are diagnosed during second decade of life and more than 50% of cases are diagnosed between the age of 13 and 19 years. We report a case in the fourth decade. The size of the lesion in various studies is commonly between 1.5 and 3 cm, but there are reports of lesions that have reached a size of 6 to 7 cm (7). Present case of large AOT measured around 6.2 X 3.1 cms in size. It is a nonaggressive epithelial tumour derived from the epithelial component of the tooth forming tissues and exhibits slow and progressive growth. But none of the associated teeth were described as morphologically defective. Thus, for the formation of lesion the disturbance must occur after odontogenesis is complete. Stafne originally suggested that the cell of origin was from epithelium entrapped in the line of embryonic fusion. The current belief is that the origin of AOT is from the odontogenic epithelium of the dental lamina complex or its remnants (8).

Clinically tumour is well circumscribed and encapsulated although sometimes it may be partly cystic. The three clinicopathological variants of Adenomatoid odontogenic tumour are intraosseous follicular, intraosseous extra follicular, and peripheral type. The intraosseous follicular type is a central lesion of bone usually found in association with an unerupted tooth and accounts for about 73% of all the AOT cases. The extrafollicular type is also a central lesion but unrelated with an impacted tooth and accounts for about 24% of all the AOT cases. Finally, the peripheral variant is a rare form attached to the gingival structures and accounts for about 3% (3). An 'extrafollicular' type AOT presents as a well-delineated radiolucent lesion. It can resemble a residual, a radicular, or a lateral periodontal cyst. It may not show a visible relation to teeth due to its deep placement. Present case represented the extrafollicular type of AOT since there was no associated impacted tooth. Erosion of the alveolar bone crest, root resorption as well as perforation of the cortical bone may also be observed radiographically as was seen in the present case (7). Small radiopaque spots or calcifications can also be an internal component of AOT. In our case the panoramic as well as the occlusal radiograph did not reveal any radiopacities. CBCT gives a better display of

the extent, border, surrounding structures and detailed intralesional content of a particular lesion with three-dimensional visualisation. CBCT, in our case, could clearly delineate the size of the lesion. The thinning and perforation of the buccal and lingual cortices could be clearly visualized. Labial displacement of the associated teeth along with the root resorption of the teeth was clearly evident in CBCT. CBCT also confirmed the unilocular nature of the lesion as well as the absence of intralesional calcifications. Although such calcifications are considered as one of the distinct features of AOT, the incidence of such radiopacities varies among different studies. Earlier studies found that approximately two-thirds of AOTs had radiopaque foci. One study later proposed that only 7.1% of cases were observed with radiopaque calcifications inside the lesion. In another study, none of the thirty-three cases had intralesional calcification (9). In our case the radiolucency was devoid of any radiopacities suggestive of calcifications. Displacement of teeth without root resorption is a commonly noted feature of this lesion (10). But in our case displacement of teeth along with root resorption was seen radiographically. Differential diagnosis of AOT includes dentigerous cyst, keratocystic odontogenic tumour, calcifying odontogenic cyst (COC) and calcifying odontogenic tumor (COT). Distinguishing AOT from dentigerous cysts is extraordinarily difficult especially when the AOT lesion is follicular and perceived as completely radiolucent (9).

Fine calcifications seen radiographically helps in differentiating an AOT from dentigerous cyst and keratocystic odontogenic tumour. Radiolucency with multiple radiopaque foci particularly when the radiolucency surrounds a portion of the root or entire tooth is suggestive of an AOT rather than a COC/COT (10). Due to the history of rapid growth, bicortical expansion and absence of calcifications, we considered unicystic ameloblastoma in the differential diagnosis. Histopathologically, AOTs consist of proliferating epithelium surrounded by a well-defined fibrous capsule. Spindle and polyhedral cells often form nodules with "duct like" cystic spaces of varying sizes seen between the nodules. Epithelial cells sometimes form small nests termed as "rosettes" or large loops connected to each other in a "plexiform" pattern. The cytoplasm is lightly stained, and oval nuclei are polarized away from the lumen (7). The treatment modality of choice is conservative surgical enucleation. Recurrence of AOT is exceptionally rare due to its encapsulated nature. Therefore, the prognosis is excellent (6,11).

CONCLUSION

Adenomatoid odontogenic tumour commonly occurs in the maxillary anterior region and associated with an impacted tooth. Reported case of AOT presented with many unusual characteristics. The large size of the lesion along with duration of only three months is suggestive of a neoplasm rather than a hamartoma. The AOT presented is in a male subject in the 4th decade while it commonly occurs in females and rarely over the age of 30 years. Radiographically, it presented with complete absence of calcifications. Root resorption, a very rare feature of AOT, was also present. All the above features thus make the case unique in presentation.

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