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CASE STUDY

PERIPHERAL ADENOMATOID ODONTOGENIC TUMOR OF THE ANTERIOR MAXILLARY GINGIVA: A RARE CASE REPORT WITH CBCT FINDINGS

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ABSTRACT

Adenomatoid Odontogenic Tumor (AOT) is a benign odontogenic tumor arising from the Odontogenic epithelium without odontogenic ectomesenchyme. Peripheral variant of Adenomatoid Odontogenic Tumour (PAOT) is relatively a rare entity which has been infrequently reported in the literature. These uncommon clinical variants of an AOT typically manifest as a soft tissue tumor of the gingiva with an identical histopathologic presentation as their intraosseous counterpart. Only fourteen cases have been adequately documented so far. In this case report we are reporting a case of PAOT of the anterior maxillary gingiva with CBCT (Cone beam computed tomography) findings which aided in better visualization of the lesion compared to conventional radiography.

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INTRODUCTION

Peripheral odontogenic tumours (POTs) are tumours that demonstrate the histologic characteristics of their intraosseous counterparts but occur solely in the soft tissue covering the tooth-bearing portion of the mandible and maxilla. These lesions are also known as extraosseous odontogenic tumours, soft tissue odontogenic tumours or odontogenic tumours of the gingiva. (Balwani Satish *et al*., 2007; Manor *et al*., 2004; Buchner *et al*., 2006; Manor *et al*., 1999) Adenomatoid Odontogenic Tumor (AOT) was first described by Dreibladt in 1907 as a pseudoadenoameloblastoma. (Lucas, 1984) In 1948 Stafne considered it as a distinct entity, although others classified it as a variant of Ameloblastoma. (Stafne, 1948) The other synonyms given for this lesion are adenoameloblastoma, adenoameloblastic odontoma, epithelial tumor associated with developmental cysts, ameloblastic adenomatoid tumor and adenomatoid or pseudoadenomatous ameloblastoma. (Stafne, 1948; Philipsen and Birn, 1969) Philipsen and Birn were the first to propose the term AOT in 1969 and expressed that it is not to be regarded as a variant of ameloblastoma because of its different behavior. (Philipsen and Birn, 1969) Phillipsen *et al* suggested three clinicotopographic variants of AOT: Follicular (pericoronal)- 73% of all cases, Extrafollicular (extracoronal) – 24% of all cases and Peripheral (extraosseous) – located in the gingival mucosa & clinically appears as a gingival fibroma or fibrous epulis and accounts for 3% of all cases. (Philipsen and Birn, 1969; Kramer and Pindborg, 1971; Philipsen *et al*., 1992) Peripheral lesions rarely are detectable radiographically but there may be slight erosion of the underlying alveolar bone cortex. One reported case demonstrated central and peripheral involvement; it could not be determined whether the bilobed lesion was primarily a gingival lesion that had eroded into the underlying alveolar bone or if a superficial, primarily intraosseous lesion had expanded out into the overlying gingiva. (Rick, 2004) The perception of internal radiopacities, particularly in cases with minimal intralesional calcification (such as the present case) becomes critical. In such situations advanced imaging like CBCT can be adopted by the radiologist in determining the extent, border, surrounding structures & detailed intralesional content in a three dimensional view. Here we present a rare peripheral AOT case in a female patient with illustration of characteristic features of the lesion on CBCT and correlation of the findings with histopathological examination.

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Case Report

A female patient aged 21years visited our department with the chief complaint of a painless swelling in the upper front tooth region since 6 months. The patient first noticed the swelling when she had discomfort while closing lip tight. Swelling was gradual in onset, slow growing, progressively increased to the present size. History revealed a similar swelling in the same region 2years back for which she visited a private dentist, following which incision and drainage was done. She was asymptomatic for 1 year until the recurrence of the swelling 6 months back for which she visited us. On extra-oral examination, no notable findings were recorded. Intraoral examination showed presence of solitary well defined swelling in the labial surface of maxillary anterior gingiva of 11, 12, roughly oval in shape measuring approximately 1X1.5cms in size. The swelling extended mediolaterally from the mesial surface of 11 to distal surface of 12 and superoinferiorly from the marginal gingiva to the depth of the vestibule (Fig 1). Loss of scalloping irt 11 was seen. Stippling was lost irt upper anteriors. Discoloration of the teeth was absent and 11 was palatally placed (Fig 2). Spacing was seen between 11, 12, 21, 22. The swelling was non-tender on palpation, with varying consistency; soft in consistency at the depth of the vestibule, and firm to hard in the middle and lower third of the swelling towards the incisal edge. Surface texture was smooth and surrounding mucosa was non tender. Grade I mobility irt 11, 12 was noted with a delayed response to electric pulp testing. The case was provisionally diagnosed as radicular cyst irt 11, 12 and a differential diagnosis of Adenomatoid Odontogenic tumor, calcifying epithelial odontogenic cyst, peripheral giant cell granuloma, peripheral ossifying fibroma was considered. Intraoral periapical radiograph and Panoramic radiograph revealed well defined solitary unilocular radiolucent lesion with internal trabeculation at the middle third of the radicular portion of 11 and 12 measuring approx. 9x7mm in size roughly oval in shape (Fig 3). Maxillary anterior occlusal radiograph revealed well-defined unilocular radiolucency with internal trabeculation. The lesion was roughly oval in shape with a corticated border measuring approx. 1.6x1cm in size extending mesiodistally from the mesial surface of 11 to distal surface of 12 and superioinferiorly from the incisal third of 11, 12 till the floor of the nasal fossa on the right side. Palatal displacement of 11 was noted (Fig 4).

For a more precise diagnosis CBCT was performed, the scan included the region of maxilla from 16 to 22. A Saucer shaped labial cortical defect was noted extending from distal surface of 21 to mesial surface of 12 in the axial section (Fig 5A). The lesion measured about 13mm x 14mm x 10mm in the coronal section (Fig 5B). A thin corticated labial outline of a soft tissue swelling along the defect was observed along with numerous small specks of calcification scattered along the periphery of the defect closer to the labial surface of the bone which was noted in the sagittal section (Fig 5C). In relation to the labial root surface of 11 there was root resorption extending close to the root canal which was separated by a thin layer of dentin. 11 was also palatally tipped. Radiologic differential diagnosis of peripheral variant of adenomatoid odontogenic tumor (AOT), calcifying odontogenic cyst (COC), keratocystic odontogenic tumor (KCOT), calcifying cystic odontogenic tumor (CCOT) was considered. Fine needle aspiration of the lesion yielded a yellow straw color fluid which on cytological examination showed few squamous cells and scanty inflammatory cells in fibrinous background. Endodontic treatment of 11, 12 were done. Surgical enucleation of the lesion was done under local anesthesia (Fig 6) by placing a trapezoidal incision extending from 13-23, & elevation of mucoperiosteal flap. Apicoectomy with apex closure using MTA retrograde filling was done irt 11. The flap closure was done using 3-0 silk suture. Enucleated tissue was sent for histopathological examination which on H& E staining showed stroma consisting of cuboidal and spindle epithelial cells arranged in solid sheets, rosette and duct like structures. Eosinophilic coagulum was seen within the duct like structures. Eosinophilic amorphous material and calcified bodies along with dentinoid material was also seen suggestive of Adenomatoid Odontogenic Tumor (Fig 7A, B). Considering the clinical, radiographic and histopathological examination, the case was diagnosed as PAOT irt 11 and 12. Patient was reviewed after 3 months; the surgical site was completely healed with normal appearing mucosa (Fig 8). Post-operative radiograph revealed new bone formation in the surgical defect (Fig 9). The patient is asymptomatic and there is no evidence of recurrence till date.

Table 1. Odontogenic cysts and tumors with internal calcifications

Calcifying odontogenic cyst
Adenomatoid odontogenic tumor
Calcifying epithelial odontogenic tumor
Ameloblastic fibrodontoma
Rarities: keratocystic odontogenic tumor, central odontogenic fibroma, radicular cyst

Fig. 1. Clinical photograph shows solitary swelling irt 11, 12 extending from the marginal gingiva to the depth of the vestibule

Fig. 2. Clinical photograph showing palatal displacement of 11

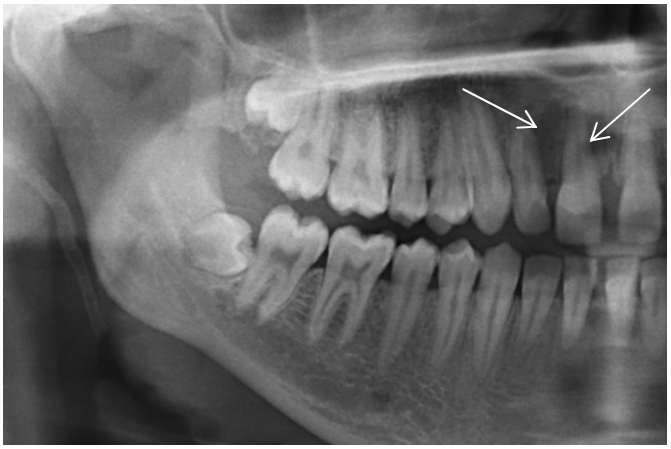

Fig. 3. Cropped Digital Panoramic radiograph of the right side showing well defined unilocular radiolucency measuring approximately 9x7mm in size irt 11 and 12

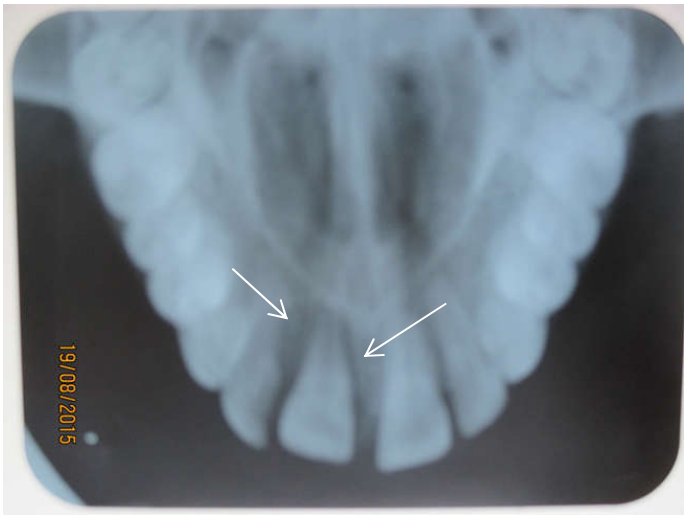

Fig. 4. Maxillary anterior occlusal radiograph showing well defined unilocular radiolucency irt 11 and 12

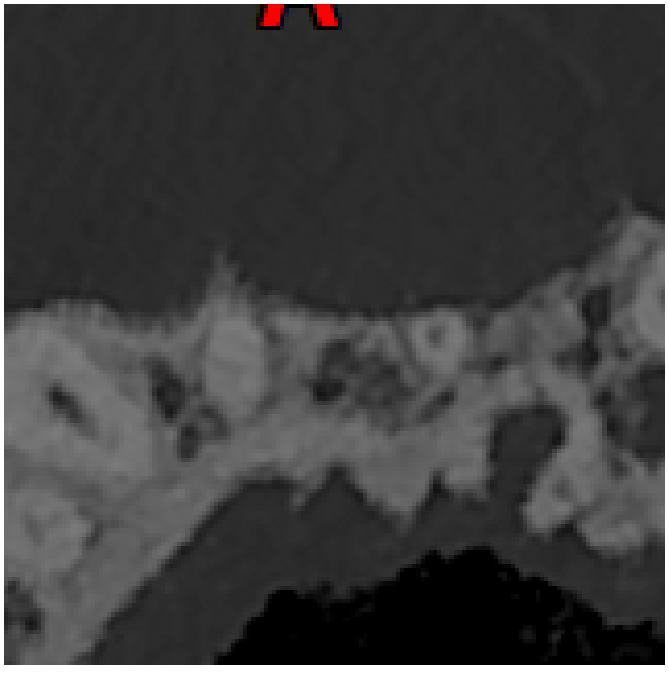

Fig. 5a.

Fig. 5b.

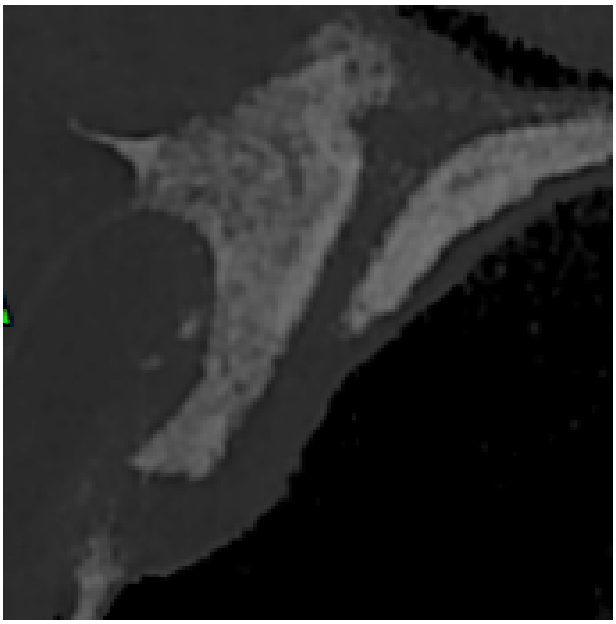

Fig. 5c.

Fig. 5. a) Axial CBCT section of the lesion shows labial cortical defect irt 11 and 12 and soft tissue extension of the lesion. b) Coronal CBCT section shows the radiopaque outline of the lesion surrounding the apex of 11 with few flecks of calcification in the internal structure. c) Sagittal CBCT section showing cortical defect irt 11 and 12 and calcification scattered along the periphery of the defect

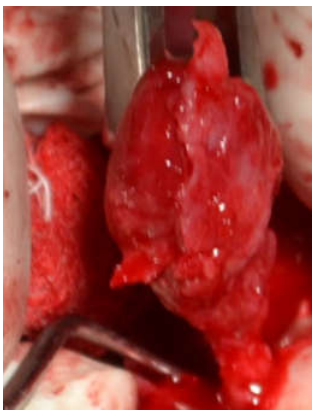

Fig. 6. Clinical photograph showing surgical enucleation of the lesion

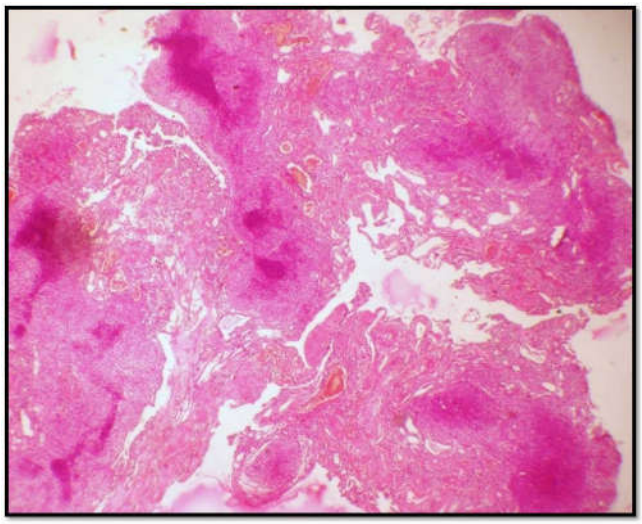

Fig. 7a.

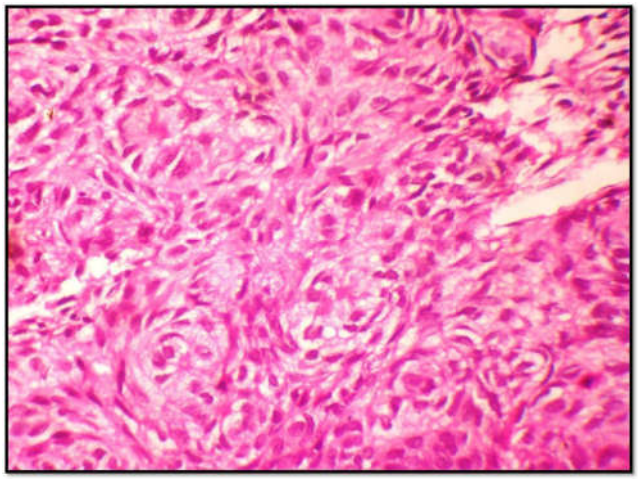

Fig. 7b.

Fig. 7. a) Histopathological picture in low magnification showing sheets of epithelial cells along with ductal pattern. b) Histopathological picture in high magnification showing sheets, nests of polyhedral cells along with ductal pattern

Fig. 8. Clinical photograph at 3 months follow up showing completely healed surgical; site

DISCUSSION

Peripheral AOT comprises between 2.3% and 4.5% of all cases of AOT; the lowest frequency of the three AOT types. (Ide *et al*., 2008) Similar to all the variants of AOT, PAOT shows a distinct male: female ratio ranging from 1:2 to 1:14.

Fig. 9. Intraoral digital radiograph taken at 3 months follow up showing new bone formation at the surgical defect

The peak incidence of the PAOT is in the second decade, with a reported mean age at presentation ranging from 11.9 to 14 years; the lowest mean age for any AOT variant. (Ide *et al*., 2008; Philipsen *et al*., 2007) The peripheral lesions are most commonly seen in the anterior maxilla, most frequently in the incisor or canine region, but have also been reported in the anterior mandible. The PAOT is the only peripheral odontogenic tumor that shares the age, gender and site predilections of its central counterpart. (Leon *et al*., 2005; Philipsen *et al*., 1992) A review of 48 well-documented cases of peripheral epithelial odontogenic tumors showed that the PAOT accounted for 12.5% of all the cases. (Buchner and Sciubba, 1987) In 2006, a review of 45 cases of peripheral odontogenic tumors collected over a 20 year period from a large biopsy service revealed no cases of PAOT. (Buchner *et al*., 2006) The clinical appearance of the PAOT is that of a painless, slow-growing gingival swelling. One reported case produced protrusion of the associated teeth (Buchner and Sciubba, 1987).

As in present case, the patient also presented with similar clinical appearance with palatal displacement of 11. The duration of the lesion may vary from months to years. (Buchner and Sciubba, 1987; Yazdi and Nowparast, 1974) The present case was peripheral variant wherein the lesion was superimposed on the labial surface of 11 and 12. In accordance with the previous reports, present case also showed female predilection and age of the patient was also favourable for PAOT and the lesion was seen in the anterior maxilla which was similar to previous reports. Radiographic appearance of PAOT may vary from normal to demonstrating evidence of superficial erosion of the underlying bone. (Bowers *et al*., 2012; Manor *et al*., 2004) Whereas, in the present case there was saucer shaped labial cortical defect associated with labial root resorption of 11 probably due to the pressure from the long standing lesion. Dystrophic calcification, believed to be dentinoid or cementum in nature, of varying amounts and patterns, may be seen in the internal structure of the lesion. The number, size and degree of calcifications present will influence the radiographic appearance of the lesion. (Regezi *et al*., 2008). Intralesional calcification with characteristic pattern was proposed as a distinctive radiographic feature of AOT. Dare *et al*. found that the radiopacities in AOT show as discrete foci having a flocculent pattern within radiolucency

even with minimal calcified deposits. (Dare *et al*., 1994) Some authors have even described the calcification as multiple minute variable-shaped calcifications or radiopaque foci, which may appear like a 'cluster of small pebbles'. (Day *et al*., 1997; Motamedi *et al*., 2005; Robert *et al*., 1995) Therefore, the capability to recognize the characteristic calcification on radiographs even in small amounts is highly meaningful for diagnosis. However, the frequency of characteristic radiopacities perceived on radiographic images varied among different AOT studies. Based on the study of hundreds of AOT cases, Philipsen *et al* reported approximately two-thirds of the intrabony AOTs possessed scattered radiopacities in the radiolucent lesions. (Philipsen *et al*., 1992) By contrast, the frequency of radiopacities in some other studies was very low. Arotiba *et al* reported that only 2 of 56 intra-osseous AOTs displayed intralesional calcifications (Arotiba *et al*., 1997), while Mohamed *et al* reported that none of 33 cases had radiopaque foci observed inside. The perception of patterned calcifications on radiographs was mainly affected by the amount of calcifications and the radiographic technique adopted (Mohamed *et al*., 2010). As an advanced imaging modality, the main advantage of CBCT is the multiplanar cross-sectional images in various orientations and three-dimensional reconstructions based on a single scan of fields of view of interest, varying from a single tooth to the whole maxillofacial area. (Jiang *et al*., 2014) CBCT is also advantageous in terms of demonstrating the detailed internal structures of lesions (eg. radiopaque calcified deposits), particularly when the calcifications are minimal or the superimposition is serious in the maxillary region. (Jiang *et al*., 2014; Becker *et al*., 2012) Moreover, CBCT provides better display of the extent and complex spatial relationship of the lesions with the surrounding structures (Jiang *et al*., 2014; Becker *et al*., 2012).

In the present case there were numerous small specks of calcification scattered along the periphery of the defect which was well appreciated in the CBCT imaging than the conventional radiographs and was beneficial in diagnosis. Previous studies of other odontogenic tumors with internal calcifications such as Calcifying cystic odontogenic tumors (CCOTs) also showed that CBCT could better demonstrate the minimal calcification than plain radiographs by avoiding the superimposition of surrounding structures. The pattern of calcified deposits of CCOT was more likely a bigger mass calcification at the bottom or to one side of the lesion. The scattered calcified foci around the crown and neck of the associated impacted tooth as well as the constant radiolucent band that might be the fibrous capsule delineating the tumor were considered as distinctive radiographic features of AOT different from CCOT. (Chindasombatjaroen *et al*., 2012) Calcifications or hard tissue deposits in KCOT are known to be rare and with the advent of CBCT even very less calcifications may be detected with ease and the calcifications are mostly seen towards the cystic wall which can be distinguish from AOT calcifications. (Naveen *et al*., 2011) Dystrophic calcifications may be observed in approximately 15% of radicular cyst (RC). A study evaluated a series of residual RC and revealed that mineralization increases with time. However, the amount of calcifications usually does not reflect radiographic images. CBCT findings of RC show great amount of hyperdense foci that causes enlargement and disruption of the cortical bone. (Lin *et al*., 2010) Calcifying odontogenic cyst (COC) show unilocular form with well-defined margin. While in 5-13% of the cases they are multilocular. They have scattered irregular sized calcification producing a variable range of opacities [salt & pepper type of patterns] which aids in differentiation from AOT (Rushton and Horner, 1997). The interrelationship between radiologic features and histologic pattern of AOT had been analysed in previous studies. (Jiang *et al*., 2014) The results indicated a positive correlation between the extent of calcifications evaluated histologically and the degree of radiopacities on radiographs. In the present case too there was positive association with the radiographic and histological features. The histology of the tumour does not differ widely; the histological appearance of all variants (follicular, extra-follicular and peripheral) is identical and shows remarkable consistency. The PAOT, like the intraosseous lesion, is usually solid, however, in this case, the lesion had a cystic component. (Shivakumar and Sahana, 2010) Unlike the intraosseous lesion, PAOTs are generally unencapsulated, although these lesions have also been reported as surrounded by a relatively thick capsule, as was observed in this case. Treatment of the PAOT involves conservative surgical excision and evaluation and appropriate treatment of an underlying bony defect if present. There have been no reports of recurrence of this lesion. (Buchner and Sciubba, 1987) We speculate that incomplete management of the lesion previously, resulted in its apparent recurrence in this case. Because of the obvious encapsulation of the lesion and histopathological and clinical evidence of complete excision in this case, recurrence is not expected.

Conclusion

The peripheral type of AOT is relatively rare compared to other types, hence meticulous clinical examination and appropriate use of advanced imaging modalities like CBCT increase the diagnostic potential over conventional radiography. In the present case, CBCT was advantageous in displaying the extent, border, surrounding structures and detailed intralesional calcifications of the lesion which was not detected in routine radiographs. Hence CBCT adds greatly to the diagnostic ability of the oral physician and aids in appropriate management.

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