



## CASE REPORT

### MANAGEMENT OF UNILOCLAR RADIOLUCENCY WITH RADIOPAQUE MASS IN ANTERIOR MAXILLA- A SYSTEMATIC APPROACH

**\*Dr. Suraj C. Bangar, Dr. Sanyukta Khairnar, Dr. Bhagyashree Jagtap, Dr. Kalyani Bhate, Dr. Santhosh Kumar, S. N. and Dr. Sushmita Mitra**

Department of Oral and Maxillofacial Surgery, Dr. D.Y. Patil Vidyapeeth,  
Dr. D.Y. Patil dental College and Hospital, Pimpri, Pune 18

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#### ABSTRACT

Swelling in the head and neck region are most commonly presented to a surgeon that may be reactionary, benign or malignant. Adenomatoid odontogenic tumor (AOT) is a well-established, benign and rare variant accounting for about 1% to 9% of all odontogenic tumors commonly occurring in young female. The report contains an unusual case of 16 years old female with cystic AOT in relation to impacted canine on right anterior maxilla, mimicking dentigerous cyst. This case report aims to highlight the clinical course, histological features and surgical management of cystic AOT. Treatment modality opted for the case was conservative with excellent prognosis and negligible chances of recurrence.

## INTRODUCTION

Odontogenic tumours are lesions derived from epithelial, ectomesenchymal and/or mesenchymal elements that are, or have been, part of the tooth-forming apparatus. The most frequent tumour was ameloblastoma (39.6%), followed by odontoma (20.1%) and the newly included keratocystic odontogenic tumor (13.8%) (Rick, 2004). Adenomatoid odontogenic tumor (AOT) is a rare, odontogenic, benign, non-invasive tumor with a slow and progressive growth accounting for about 1% to 9% of all odontogenic tumors (Philipsen and Reichart, 1998). According to recent WHO classification of odontogenic tumors, AOT is defined as being composed of odontogenic epithelium in a variety of histo-architectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth (Philipsen *et al.*, 2007). Philipsen and Birn proposed the widely accepted and currently used name adenomatoid odontogenic tumor, a term that was adopted by the first edition of the World Health Organization classification of odontogenic tumors in 1971 (Philipsen and Reichart, 1998; Philipsen *et al.*, 2007). AOT is usually found in young patients in second decade of life (Shetty *et al.*, 2005).

This case presentation highlights the diagnosis and management of AOT mimicking as a dentigerous cyst both clinically and radiographically. In 1915, Harbitz reported the cystic presentation of AOT as "cystic adamantoma" (Philipsen and Reichart, 1998). The histogenesis of several variations of AOT is controversial (Reichart and Philipsen, 2004). In this case, significant radiographic finding helped to exclude the dentigerous cyst from the final diagnosis.

## CASE REPORT

A 16 years old female patient, presented with an intraoral swelling and pain in the upper right front region of the jaw since 6 months. Patient noticed a painless swelling six months back in the right front region of upper jaw. The swelling began as a small peanut and gradually increased to the present stage. Swelling was associated with pain since last 1 month. Pain was continuous and dull aching in nature and it subsided with medication. On inspection, extraorally there was a diffuse swelling of the right cheek region with obliteration of nasolabial fold. (Figure 1) Intraorally a solitary, ovoid swelling was present, which was extending from right maxillary lateral incisor to first premolar obliterating the labial vestibule (Figure 2). On palpation, swelling was firm, hard and non-tender with ill-defined borders. Egg shell cracking sound was appreciated on palpation.

\*Corresponding author: Dr. Suraj C. Bangar,  
Department of Oral and Maxillofacial Surgery, Dr. D.Y. Patil Vidyapeeth, Dr.  
D.Y. Patil dental College and Hospital, Pimpri, Pune 18, India.



Figure 1. Diffuse swelling of the right cheek region



Figure 2. Swelling in right maxillary cheek regionalveolus and missing canine



Figure 3. Swelling bucco-palatal bony expansion with missing 13, 14, 15 and retained deciduous molar

The overlying mucosa was intact with an edentulous space distal to the lateral incisor. The canine, 1<sup>st</sup> and 2<sup>nd</sup> premolar was missing from the dentition. Mobile as well as over retained deciduous second molar (E) and grossly carious teeth (36, 46) were present in the arch (Figure 3). Radiographic examination (Orthopantomogram) revealed inverted pear shaped, well demarcated unilocular radiolucency with radio opaque masses, displaying smooth corticated borderpericoronal to the impacted canine (13).



Figure 4. Pre-operative panoramic radiograph (OPG) revealing a well-defined radiolucent lesion around the impacted 13 and unerupted 14, 15 in the right maxilla



Figure 5. CBCT imaging reveals inverted pear shaped radiolucency with radiopaque masses and approximation with maxillary sinus and nasal floor

Roots resorption of 1<sup>st</sup> premolar (14) and divergence with respect to lateral incisor and both premolars (14, 15) was appreciated on radiograph. (Figure.4). Close relation with respect to the nasal floor and maxillary sinus was seen on CBCT examination (Figure 5). On the basis of above clinical and radiographic findings differential diagnosis considered was dentigerous cyst, adenomatoid odontogenic tumor, calcifying odontogenic cyst, calcifying odontogenic tumor, keratocystic odontogenic tumor and unicystic ameloblastoma. Association of an impacted tooth, radiolucency surrounding the tooth portion leads towards provisional diagnosis of dentigerous cyst. Another provisional diagnosis concluded was globulomaxillary cyst based upon radiographic findings of inverted pear shaped radiolucency. The extra-follicular type is not associated with an unerupted tooth and well-defined, inverted pear shaped unilocular radiolucency is found around the roots of erupted permanent teeth. These findings may lead to preoperative diagnosis of globulomaxillary cyst. Surgical plan decided was complete excision of the lesion along with extraction of involved teeth, (12, 13 and 15) without bone graft under general anesthesia. After the preoperative investigations, pre-anesthetic fitness, valid, written, informed and signed consents were obtained from patient. After naso-endotracheal intubation and scrubbing, painting and draping was done. Local anesthetic with adrenaline (1:200000) was infiltrated from 12 to 16 region. Crevicular incision was made, from 15 extending to 21 along with vertical releasing incision made distal to 15. Triangular full thickness Full thickness mucoperiosteal flap reflection was done. The lesion site was identified and exposed. A bony window was created on the expanded buccal cortical bone using no.8 round bur and 701 straight fissure bur with a postage stamp method. (Figure 6) Bony window was expanded further with bone ronger and lining of the lesion was exposed.



Figure 6. Exposure of lesion and bony window preparation with postage stamp method

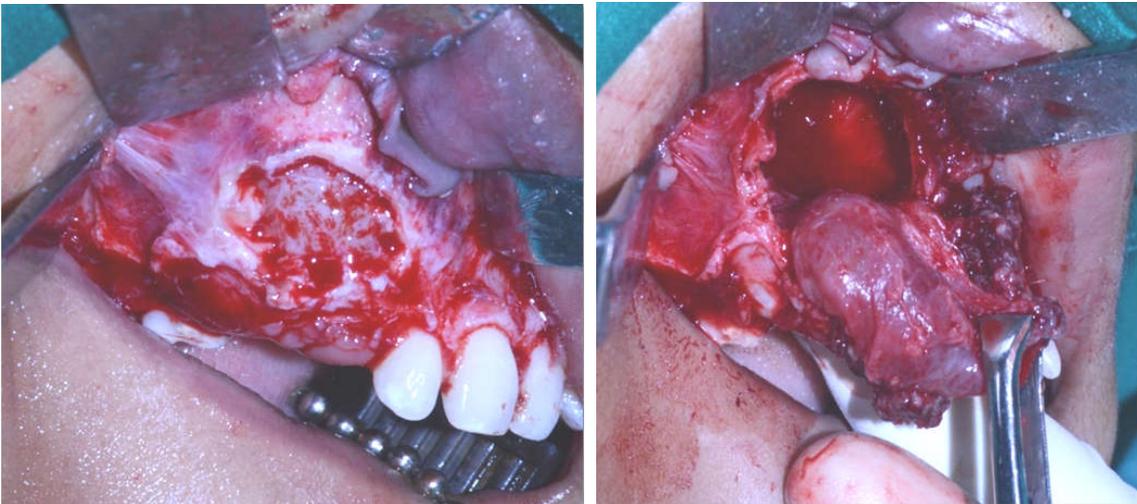
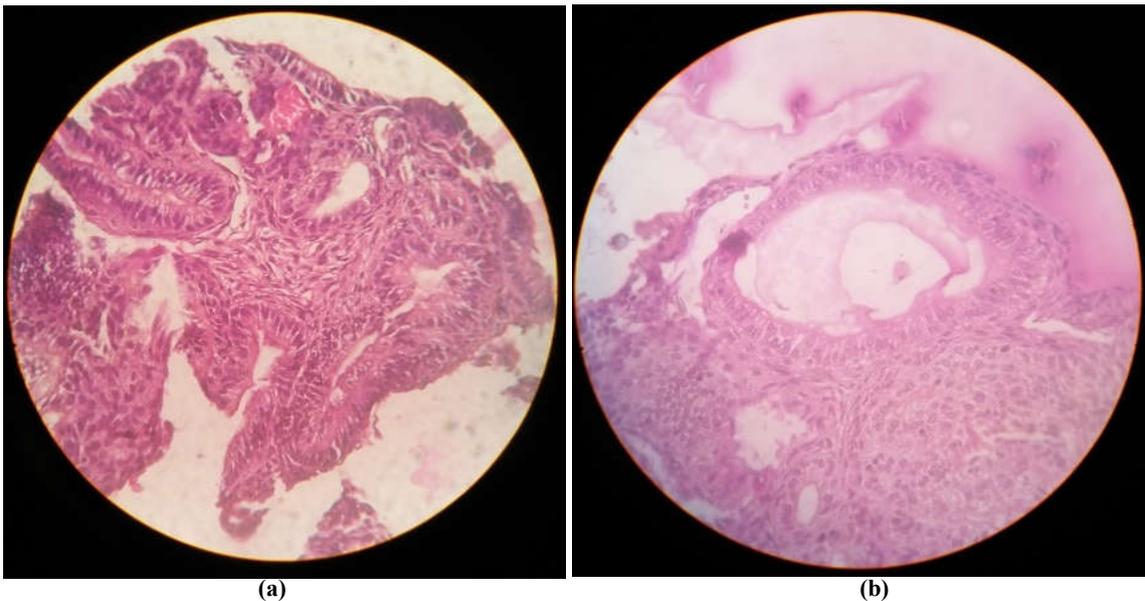


Figure 7. Excision of tumor mass lining along with impacted canine



(a)

(b)

Figure 9(a). Photomicrograph showing double layered of cuboidal and columnar cells arranged in form of nest and rosette pattern. (Hematoxylin and Eosin stain; Total magnification x100)

Figure 9(b). Photomicrograph showing cells arranged in ductal pattern with eosinophilic coagulum. (Hematoxylin and Eosin stain; Total magnification X400)



**Figure 10. Postoperative photograph with reduced swelling**

The lesion was enucleated completely in toto with embedded I3 and sent for histopathological examination (Figure. 7). Extraction was done with 12 and 15. Debridement of the bony cavity done and hemostasis achieved. Primary closure was done with 3-0 vicryl suture material. On histopathological examination, eosin and hematoxylin stained section showed two layers of cuboidal columnar cells arranged in form of nest and rosette pattern. Solid areas showed duct like pattern and whirling of these tumor cells is evident at places where as strands of tumor cells with pale eosin material. Few areas showed basophilic structures suggestive of calcification. A dense connective tissue capsule is noted with numerous extravasated RBCs. Thus, histopathological examination confirmed the diagnosis of AOT (Figure 9 a, b). Patient was kept on intravenous antibiotics and analgesic immediately postoperatively and discharged after 7 days with oral medications. On follow up visits, the swelling was reduced with satisfactory healing. (Figure 10)

## DISCUSSION

The Adenomatoid Odontogenic Tumor is a benign tumor that is keen to involve the anterior region of the maxillary bones, with a larger number of cases in females, in their second decade of life (Philipsen and Reichart, 1998). The origin of adenomatoid odontogenic tumours is controversial. Some believe that they originate from the odontogenic epithelium of a dentigerous cyst. In addition to the anterior maxilla, the tumor has been reported in other areas of the jaw, such as the angle of the mandible. Therefore, dental lamina remnants likely represent the progenitor cells for this benign odontogenic tumour. According to this hypothesis, the lesion grows (sometimes while forming a cystic space) next to or into a nearby dental follicle, leading to the envelopment theory (Philipsen *et al.*, 1992). The adenomatoid odontogenic tumour (AOT) is now recognized to consistently represent a lumen lined with a specific type of epithelial proliferation arising from Hertwig's epithelial root sheath. Conversely, in past which was identified as an adenomatoid odontogenic cyst (AOC), not a true neoplasm or hamartoma. The adenomatoid odontogenic cyst (AOC) is a cystic hamartoma arising from odontogenic epithelium (Marx *et al.*, 2003). Immunohistochemical studies report that the slow growth, its benign character and low tendency to recur are clearly related to the low cellular proliferation observed on carrying out immunostaining for the antigen.

AOT has been known for its varied histo-architectural patterns. There has always been a controversy regarding the true nature of this lesion. It was considered to be a hamartoma clinically, due to its limited size and lack of recurrence (Rick, 2004; Marx *et al.*, 2003). Philipsen and Birn proposed the name AOT (1969) and suggested that it should not be regarded as a variant of ameloblastoma because of its different behaviour (Philipsen and Reichart, 1998; Philipsen *et al.*, 1992). AOT is also called "two-thirds tumour," because two-thirds occur in young females, two-thirds of these cases occur in the maxilla, two-thirds of these tumors are associated with unerupted teeth and two-thirds occur in canines (Marx *et al.*, 2003). AOT is divided into 3 variants by Philipsen *et al.* The follicular type (accounting for 73% of cases), which has a central lesion associated with an embedded tooth as seen in this case presentation. The extra follicular type (24% of case), which has a central lesion and no connection with the tooth. The peripheral variety (3% of cases) is which occurs primarily in the gingival tissue of tooth-bearing areas. Association of an impacted tooth, radiolucency surrounding the tooth portion leads towards provisional diagnosis of dentigerous cyst. Dentigerous cyst encloses only the coronal portion of the impacted tooth with the radiolucency attached at the CEJ, whereas in AOT the radiolucency usually surrounds both the coronal and radicular aspects of the involved tooth (Reichart and Philipsen, 2004). Globulomaxillary cyst can also be a differential diagnosis because of the site and appearance clinically and radiologically (Philipsen *et al.*, 2007). Findings of inverted pear shaped radiolucency. The extra-follicular type is not associated with an unerupted tooth and well-defined, inverted pear shaped unilocular radiolucency is found around the roots of erupted permanent teeth. These findings may lead to preoperative diagnosis of globulomaxillary cyst. The tumor is associated with unerupted teeth, frequently canine and lateral incisors. Irregular root resorption is seldom reported (Dayi *et al.*, 1997; Mendis *et al.*, 1990). It is more common in maxillary canine and premolar region. All variants of AOT are well encapsulated and show an identical benign behaviour. Conservative surgical enucleation or curettage is the treatment of choice with only rare recurrence (Rick, 2004).

## Conclusion

All variants of AOT are well encapsulated and show an identical benign behaviour. Conservative surgical enucleation or curettage is the treatment of choice with only rare recurrence. The patient describe in this case report is healthy without recurrence and is under follow up after local excision. Thus, AOT must be considered in the differential diagnosis for radiolucent lesion with radiopaque masses in anterior maxillary region for young adult patients.

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