

# Atypical Presentation of Extra-follicular Adenomatoid Odontogenic Tumor of Anterior Maxilla: A Clinical Predicament

Punyasloka Pati<sup>1</sup>, Tribikram Debata<sup>1</sup>, Bijay Kumar Das<sup>2</sup>, Surya Narayan Das<sup>3</sup>

<sup>1</sup>Post-graduate student, Department of Oral Pathology and Microbiology, Sriram Chandra Bhanja Dental College, Cuttack, Odisha, India, <sup>2</sup>Professor, Department of Oral Pathology and Microbiology, Sriram Chandra Bhanja Dental College, Cuttack, Odisha, India, <sup>3</sup>Associate Professor, Department of Oral Pathology and Microbiology, Sriram Chandra Bhanja Dental College, Cuttack, Odisha, India

## Abstract

The adenomatoid odontogenic tumor (AOT) is a hamartomatous proliferation of odontogenic epithelium with slow growth potential constituting around 3% of all odontogenic tumors. The tumor has a more predilection for females in the second decade of life, involving the maxilla more commonly. The tumor present in three variants: Intra-follicular (73%), extrafollicular (24%), and peripheral (3%). We report a case of extra-follicular AOT in an 11-year-old male patient presenting with firm, painless swelling on the right anterior maxilla with respect to 11-12 tooth region without any relevant dental history leading to a provisional diagnosis of the developmental maxillary cyst. Fine needle aspiration revealed a benign neoplasm and histopathological examination along with radiographic correlation confirmed the diagnosis of an extra-follicular AOT of anterior maxilla.

**Key words:** Adenomatoid odontogenic tumor, Extra follicular, Fine needle aspiration cytology

## INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is an uncommon, expansile, hamartomatous and asymptomatic benign non-invasive lesion of odontogenic origin that was first described by Driebaldt in 1907 as a pseudo-adenameloblastoma.<sup>1</sup> After some terminological controversy, the name "AOT" was first proposed by Philipsen and Birn in 1969,<sup>2</sup> and adopted by the World Health Organization (WHO) classification of odontogenic tumors in 1971.<sup>3</sup> At present, it is recognized as AOC.<sup>4</sup> Sometimes it has been referred to as two-thirds tumor.<sup>5</sup>

There are three clinical variants such as follicular variant (F) is intraosseous associated with impacted and displaced tooth; extra-follicular variant (Sub type-E1, E2, E3, E4) is within the bone but not associated with unerupted tooth

rather in between erupted tooth mimicking a radicular cyst or lateral periodontal cyst; peripheral (epulis-like) variant (P) exhibits a periodontal bone defect or ectopic growth.<sup>6</sup>

Here, we reported a case of extra-follicular AOT of the maxilla, presenting as a maxillary cyst with the diagnostic challenge.

## CASE REPORT

An 11-year-old male patients presented with a painless swelling over the upper anterior region with respect to 11-12 tooth region for 3 months. Swelling was gradual in onset with the displacement of adjacent tooth with time and reach to the present condition. There was no history of dental caries, pus discharge, and tooth mobility. There was no relevant medical history, and routine blood investigation was inconclusive.

On extra-oral examination revealed slight elevation of right upper lip region with minor obliteration of nasolabial fold, no sign of inflammation, and no lymphadenopathy. Intraoral examination showed solitary bulging from upper alveolus and attached gingiva measuring about 3 × 2 cm

Access this article online



www.ijss-sn.com

Month of Submission : 06-2016

Month of Peer Review : 07-2016

Month of Acceptance : 08-2016

Month of Publishing : 08-2016

**Corresponding Author:** Dr. Tribikram Debata, Department of Oral Pathology and Microbiology, Sriram Chandra Bhanja Dental College, Cuttack - 753 007, Odisha, India. Phone: +91-9938723721. E-mail: Tribikramdbt@gmail.com

with respect to 11-12 tooth region with palatal extension and thinning out of the labial cortical plate. There was displacement of a lateral and central incisor with remains vital. On palpation, swelling was diffuse, soft to firm in consistency, non-tender, non-pulsatile, non-fluctuant, and slightly compressible and showed no evidence of discharge on digital pressure (Figure 1a).

Intra oral periapical radiograph and orthopantomogram showed well-defined unilocular radiolucency in the interdental area with the destruction of alveolar bone with respect to 12-11 tooth region and loss of lamina dura of 12 teeth only (Figures 1b and c).

From clinico-radiological evidence leads to a provisional diagnosis of the developmental maxillary cyst.

Fine needle aspiration cytology (FNAC) of that lesion yielded clusters and sheets of basaloid cells with scanty to moderate cytoplasm having round-oval benign nuclei with fine chromatin and indistinct nucleoli in a mucoid matrix and peripheral palisading also noted, imprinting a cytopathological diagnosis of benign neoplasm possibly of ameloblastoma or AOT or basal cell adenoma (Figures 2a and b).

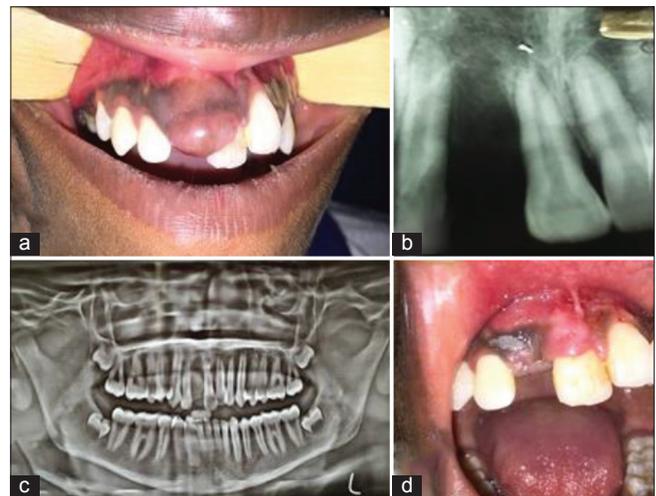
Excisional biopsy was advised and gross specimen showed grayish black solitary nodule with a cystic cavity having whitish glistening proliferating mass filled up the cavity (Figure 2c). Microscopic examination revealed proliferative epithelium arranged in whorls, nests and ducts surrounded by columnar, cuboidal cells having eosinophilic material in the center within a loose connective tissue stroma surrounded by thick fibrous capsule (Figures 2d-f). The final diagnosis was made as AOT extra-follicular type (E2 type). The present case had uneventful healing with asymptomatic after 1-month follow-up (Figure 1d). The patient was advised for orthodontic consultation for the management of the tooth spacing.

## DISCUSSION

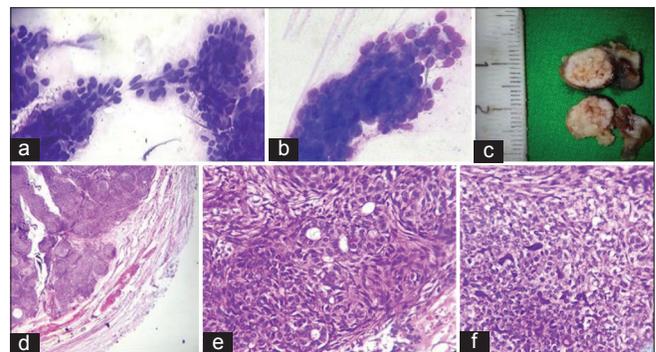
AOT is a slow growing odontogenic tumor with variable growth potential.<sup>7</sup> It occurs typically in young persons in the second or third decades with female predominance in a global incidence of 1.9:1 and for Asian 2.3: 1.<sup>8</sup> 76% of cases found in the anterior part of the jaw with a marked maxillary preference of 4.5: 2.5<sup>9</sup> and commonly located in the lateral incisor, canine and premolar region. The present case was a young male patients presented as an unusual slow growing mass over right anterior maxilla in between central and lateral incisor.<sup>7</sup>

It is frequently encountered as a painless intraosseous lesion with impacted canine.<sup>4</sup> However, it may rarely occur in a normally erupted dentition as extra-follicular type and uncommonly as a peripheral type.<sup>7</sup> The present case was not associated with impacted tooth and intra-radicular tumor location with roots divergence confirmed the diagnosis of extra-follicular E2 Type.

Radiologically, the tumor is well demarcated unilocular radiolucency, displacement of the adjacent tooth with least root resorption and sprinkle calcification which is not always sufficient to produce radiopacity.<sup>4</sup> It is always misdiagnosed as dentigerous cyst, fissural cyst, lateral periodontal cyst, radicular cyst, and nasopalatine cyst in accordance to clinico-radiological interpretation.<sup>7</sup>



**Figure 1: (a) Solitary diffuse swelling involving vestibule and attached gingiva with respect to 11-12 tooth region. (b and c) intraoral periapical x-ray and orthopantomogram showing expansile irregular destruction interdental bone and alveolar bone 11 with respect to 12 tooth region. (d) Post-treatment intraoral photograph**



**Figure 2: (a and b) Cytological smear (Leishman's, Giemsa  $\times 400$ ): Clusters and sheets of basaloid cells in a mucoid matrix. (c) Gross specimen. (d, e, and f) Histopathology (H and E stain  $\times 100$ ,  $\times 400$ ,  $\times 400$ ): Encapsulated tumour solid nodular mass, proliferative epithelium arrange in whorls, nests and ducts surrounded by columnar, cuboidal cells having eosinophilic material**

Extra-follicular AOT is often misdiagnosed by clinician due to low incidence and uncommon presentation of the disease and should be confirmed by histopathological examination. According to a review of literature FNAC of AOT has not been practiced much. In our case, FNAC not only ruled out the cystic lesion but also suggested to a cytological diagnosis of benign neoplasms - such as basal cell adenoma, ameloblastoma, and AOT - which are rarely found in this location. This finding prompted us to explore the cytological diagnosis, and the patient was advised biopsy in follow.

The histological features in our case were consistent with the histopathological criteria defined by the WHO.<sup>10</sup> This case was not only a diagnostic challenge but also gives an idea of the cytological imprint of AOT. The histogenesis of AOT is unknown and thought to arise from odontogenic epithelium, remnant of dental lamina, Hertwig epithelial root sheath.<sup>4</sup>

Conservative surgical enucleation and curettage of the lesion are the treatment of choice.<sup>8</sup> Recurrence of the tumor is very rare with an excellent prognosis.

## CONCLUSION

Extra-follicular AOT of the maxilla is a rare occurrence posing a diagnostic challenge for clinician embodying clinico-radiological features is those mimicking with many

odontogenic and inflammatory cysts. The final diagnosis of the AOT was made after histopathological examination although FNAC of the lesion in the given case has given a cytological inscription. AOT must be considered in the differential diagnosis of corticated radiolucency with or without small radiopaque foci, especially among young adults even in the absence of an impacted tooth.

## REFERENCES

1. Batra P, Prasad S, Parkash H. Adenomatoid odontogenic tumour: Review and case report. *J Can Dent Assoc* 2005;71:250-3.
2. Philipsen HP, Birn H. The adenomatoid odontogenic tumour. Ameloblastic adenomatoid tumour or adeno-ameloblastoma. *Acta Pathol Microbiol Scand* 1969;75:375-98.
3. Swasdison S, Dhanuthai K, Jaikittivong A, Philipsen HP. Adenomatoid odontogenic tumors: An analysis of 67 cases in a Thai population. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:210-5.
4. Robert ME, Dinae S. *Oral and Maxillofacial Pathology: A Rational for Diagnostic and Treatment*. 2<sup>nd</sup>ed. Chicago: Quintessence Publishing Co., Inc.; 2012. p. 639-4.
5. Neville WB, Allen MC. *Oral and Maxillofacial Pathology*. 3<sup>rd</sup> ed. Noida: Reed India Elsevier Limited; 2009. p. 713.
6. Philipsen HP, Srisuwan T, Reichart PA. Adenomatoid odontogenic tumor mimicking a periapical (radicular) cyst: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:246-8.
7. Lucas RB. *Pathology of Tumours of the Oral Tissue*. 4<sup>th</sup> ed. Edinburgh, Scotland: Churchill Livingstone; 1984. p. 61-5.
8. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumor: Biologic profile based on 499 cases. *J Oral Pathol Med* 1991;20:149-58.
9. Mars M. Adenomatoid odontogenic tumour (adenoameloblastoma). A case report. *Br Dent J* 1978;144:45-6.
10. Kramer IR, Pindborg JJ, Shear M. *Histological Typing of Odontogenic Tumours*. Berlin: Springer Science & Business Media; 1992.

**How to cite this article:** Pati P, Debata T, Das BK, Das SN. Atypical Presentation of Extra-follicular Adenomatoid Odontogenic Tumor of Anterior Maxilla: A Clinical Predicament. *Int J Sci Stud* 2016;4(5):261-263.

**Source of Support:** Nil, **Conflict of Interest:** None declared.