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Case Report

A proposed treatment algorithm for adenomatoid Odontogenic Tumor- A tumor in disguise of a cyst

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ABSTRACT:
Most odontogenic cysts or tumors are intraosseous, causing the tooth and bone to resorb; but, in some instances, they may be extraosseous and gradually growing.Adenamatoid Odontogenic Tumor (AOT) is a tumor of odontogenic epithelial origin.The clinical characteristics of AOT include painless bone enlargement, impacted teeth and facial asymmetry caused as a result of the lesion's growth. Adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium, and teratomatous odontoma are other synonyms for AOT.We report on a female patient, age 15, who was first diagnosed with a cyst but, following an excional biopsy, was found to have an AOT.
Keywords: tumor, odontogenic tumor, treatment algorithm

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INTRODUCTION

Odontogenic cyst or tumors are mostly intraosseous causing bone and tooth to resorb, but in rare cases it might be extraosseous and slow growing in nature. Mostly, patient go undiagnosed due to slow growth and asymptomatic nature of the lesion. Patients generally report at a later stage with severe bone loss & mobility of teeth prognosis of the affected region resulting in loss of teeth at an early age.

Adenamatoid Odontogenic Tumor (AOT) is a tumor of odontogenic epithelial origin. Described firstby Steensland in 1905 andDreibladt, in the year 1907 coined the term as pseudo ameloblastoma. It usually presents a slow-progressing and nonmalignant epithelial tumor that is usually benign in nature. [1]

World Health Organization classification in 1971 based on Kalia et al. proposal, has defined AOT as “a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue”.[2]

The etiopathogenesis of AOT is still unclear and controversial. AOT is considered as a developmental outgrowth or a hamartoma while others consider it as a major neoplastic growth of odontogenic epithelium. [3.4]

The Clinical features of AOT—associated with impacted teeth or in absence, painless, bony expansion, facial asymmetrydue to growth of the lesion. Various synonyms for AOT include eadenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium, or teratomatous odontoma.[1]

We report a 15-year-old female patient, provisionally diagnosed as cyst which turned out to be an AOT after excional biopsy.

CASE REPORT

A 15 year old Female patient reported to the department of Oral and Maxillofacial Surgery with a chief complaint of a swelling in her upper front tooth region for the past 2 years. The swelling had gradually increased in size over the last 2 years and was non tender to palpation the patient elicited no signs of pain

or discomfort. The patient was otherwise healthy and did not report with any other complaints.

On detailed examination extra orally there was mild facial asymmetry present in relation to right anterior maxilla and upon intra oral examination an ovoid swelling was seen in relation to right maxillary lateral incisor (12) and right maxillary canine (13) that measured approximately 3cm* 5 cm, the swelling extended from the mucogingival junction of the right maxillary lateral incisor to the crest of the alveolus of the right maxillary canine.

Tenderness was absent and the swelling showed signs of fluctuancy and non transilluminant and non pulsatile. Radiographic evaluation was done initially using and INTRA ORAL PERIAPICAL RADIOPGRAPH (IOPA) in relation to the region of interest which showed the presence of radio opaque and radiolucent structuresin relation to 12,13. IOPA also revealed a displaced 13 and a well circumscribed radiolucent lesion in relation to 12, 13. An occlusal radiograph was then taken to determine the position of 13 which also revealed a radiolucent lesion with a dense sclerotic border. After obtaining both written and verbal consent from the patient under Local Anaesthesia Fine Needle Aspiration Cytology (FNAC) was carried out which yielded in a straw colored fluid aspirate?

Based upon the history of the presentation along with the clinical and radiographic examinations we came to a provisional diagnosis of an Radicular Cyst with a differential diagnosis of AOT,Fibrous Dysplasia. Treatment plan was formulated and discussed with the patient. Ennculeation of the lesion was planned under General Anesthesia along with extraction of the displaced canine due to the poor prognosis of the tooth.

Under Aseptic conditions General Anesthesia was induced and ennucleation of the lesion was done along with extraction of the offending tooth. Histopathological examination revealed the lesion to be Adenamatoid odontogenic Tumor (AOT) Follicular type.Patient was placed on post operative follow up for a period of 6 months and it was uneventful in nature. Functional rehabilitation is planned to be done using a fixed partial denture in relation to 12-14.

DISCUSSION

Adenamatoid odontogenic tumor- an epithelial origin also termed as ‘two-thirds tumor’ because of its higher incidence of reportedoccurrences in the maxilla among young women. Almost two -thirds of the cases are associated with impactedmaxillary canines.[5]. It is the fouth most common odontogenic tumor accounting for upto 7 percent of cases of odontogenic tumor.They are usually benign slow growing tumor with better prognosis following surgical resection with minimal reports of recurrences.

AOT has three variants clinic-pathologically they include follicular variant,extra follicular variant and Peripheral type.

Whether AOT is a cyst, tumor or a hamartoma is still a matter of debate. Some authors, like Marx RE et al, Stern D et alconsider it as a true benign, non-aggressive, non-invasive neoplasm while others consider itas a hamartomatous odontogenic growth. In the recent case reports, authors had categorisedit as a cyst based on histopathologic examination exhibiting characteristics of cystic lumen, lining andconnective tissue capsule. Marx & Stern proposed the term adenomatoid odontogenic cyst and considered it to be a cyst, that has a hamartomatous intraluminal proliferation ofepithelial cells derived from Hertwig’s epithelial root sheath & these cells fill the lumen &give the impression of a solid tumor[6]

The slow growing nature of the tumor with intraosseus spread results in marked resorption of bone with teeth displacement leading to extraction. In mostcases, the asymptomatic representation of the tumor delays early intervention for surgical management. In our experience based on clinical, radiological features and histopathological findings, we have proposed a diagnosis-based treatment algorithm for easier differentiation from a cystic lesion.

WHEN TO SUSPECT AOT?

CLINICAL FEATURES:

1. INTRAORAL/EXTRAORAL SWELLING
2. ASYMPTOMATIC
3. INTRAOSSEOUS GROWTH
4. SLOW GROWING
5. PREDILICTION FOR ANTERIOR MAXILLA
6. CORTICAL EXPANSION
7. ASSOCIATED TOOTH DISPLACEMENT
8. OBLITERATION OF NASOLABIAL FOLD

RADIOGAPHIC DIAGNOSIS

IOPA- RADIOLUCENT LESION WITH/WITHOUT A DISPLACED TOOTH

OPG-RADIOLUCENT SWELLING WITH DIFFUSE BORDER

OCCLUSAL RADIOGRAPH-RADIOLUCENT LESION WITH DIFFUSE SCLEROTIC BORDERS

CBCT- MULTIPLANAR CROSS SECTIONAL VIEWS OF INTREST WITH DEMONSTATION OF INTERNAL STRUCTURES SUCH AS CALCIFIED DEPOSITS WITH A BETTER DISPLAY OF EXTENT OF LESION WITH APPROXIMATION TO VITAL STRUCTURES

BIOPSY

FNAC- USUALLY PROTIENCEOUS STRAW COLOURED FLUID WITH POSSIBILITY OF RBC WBC INTERSPERCED WITH POLYMORPHONUCLEAR CELLS AND MACROPHAGES. PROTIEN CONTENT MAYBE LESS AND IS USUALLY NOT OF CONCLUSIVE EVIDENCE.

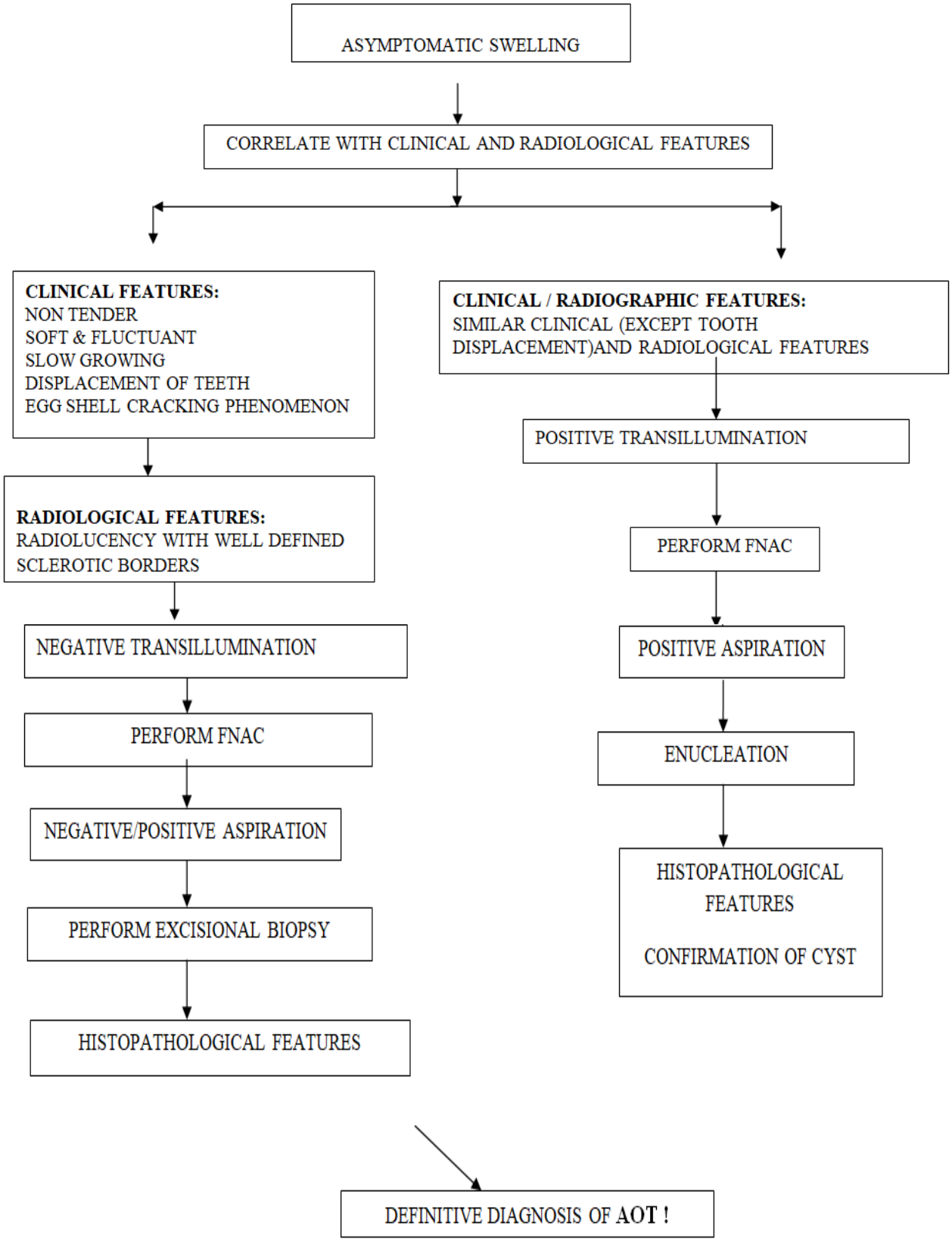
EXCISIONAL BIOPSY;

IN TOTO EXCISION OF LESION TO BE DONE WITH ENNUCLEATION

HISTOPATHOLOGICAL FINDINGS:

AOT IS MADE UP OF EPITHELIAL CELLS ARRANGED IN STRANDS OF SPINDLE-SHAPED CELLS, EPITHELIAL SPHERES/WHORLS, AND CUBOIDAL EPITHELIAL CELLS ARRANGED IN DUCT-LIKE STRUCTURES WITH OR WITHOUT CALCIFICATIONS

TREATMENT ALGORITHM FOR ASYMPTOMATIC SWELLING



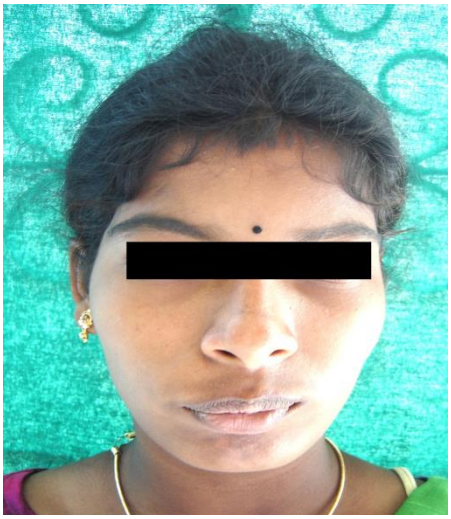


FIGURE 1: FRONTAL PROFILE SHOWING MILD FACIAL ASSYMETRY IN RIGHT ANTERIOR MAXILLARY REGION

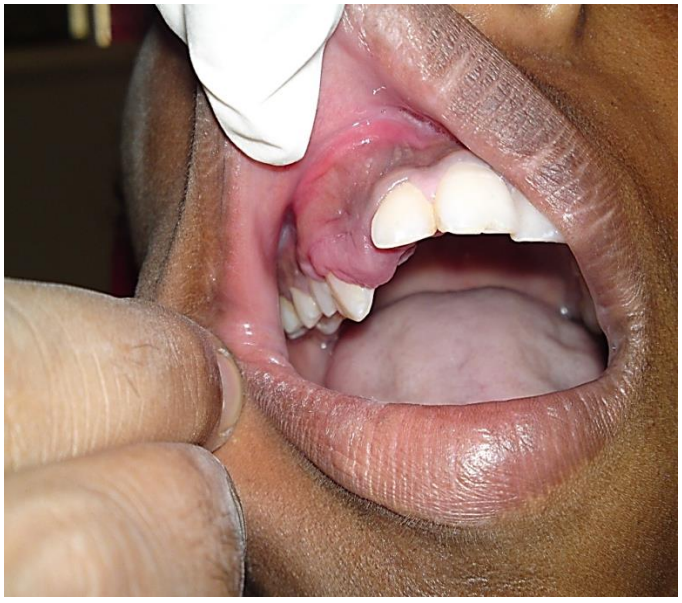


FIGURE 2: INTRA ORAL OVOID SWELLING PRESENT IN RIGHT ANTERIOR MAXILLA

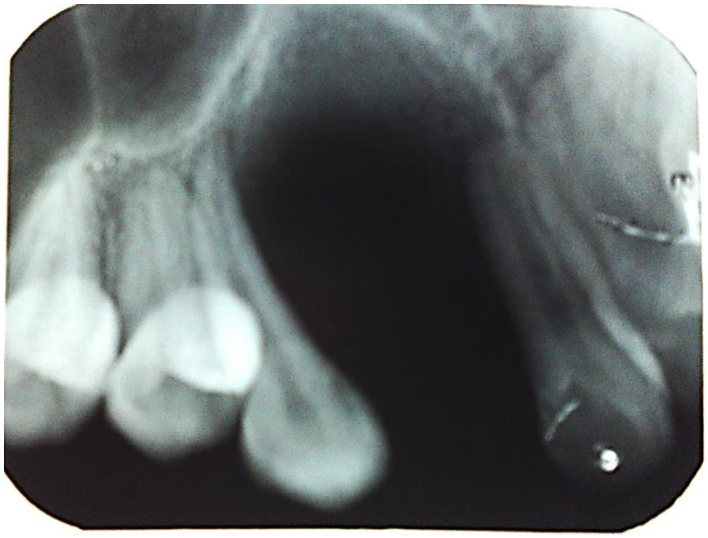


FIGURE 3; IOPA IN RELATION TO 13,14 SHOWED PRESENCE OF WELL CIRCUMSCRIBED RADIOLEUCENT LESION ALONG WITH DISPLACEMENT WITH 13.

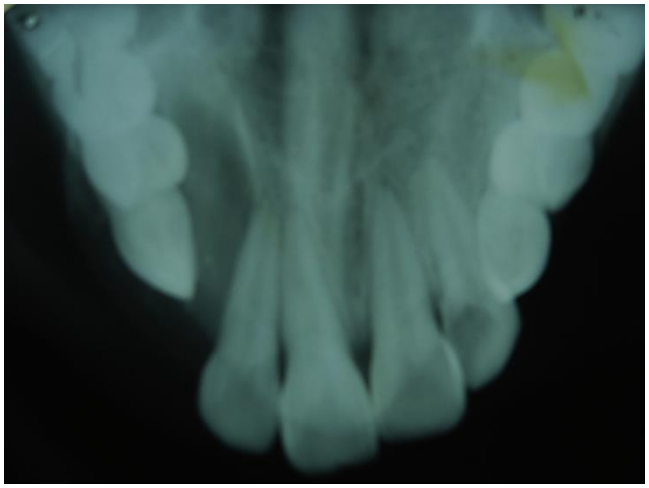


FIGURE 4: OCCLUSAL RADIOGRAPH SHOWING BICORTICAL EXPANSION WITH SCLEROTIC BORDERS

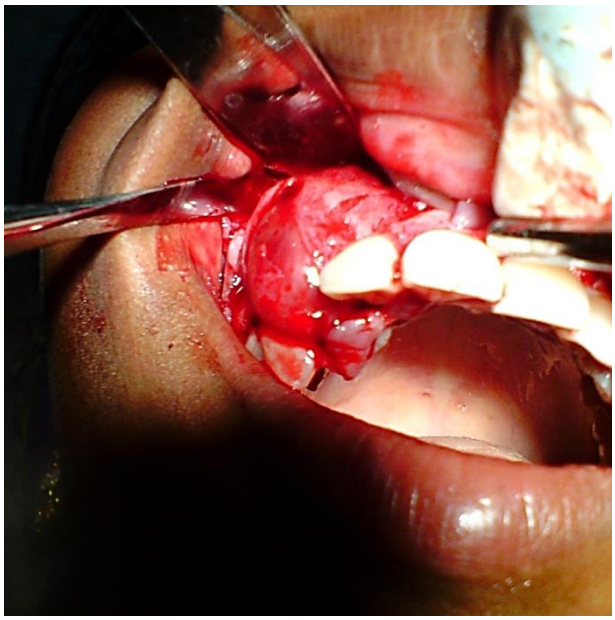


FIGURE 5&6: INTRA OPERATIVE EXPOSURE AND EXCISION OF LEISION

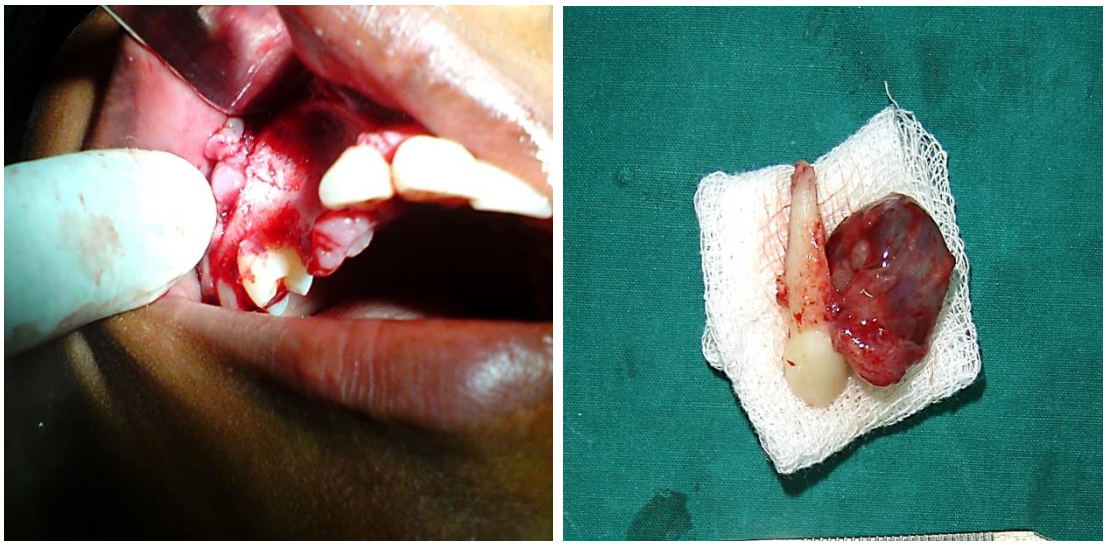


FIGURE 7: EXTRACTED SPECIMEN FOR HISTOPATHOLOGICAL EXAMINATION

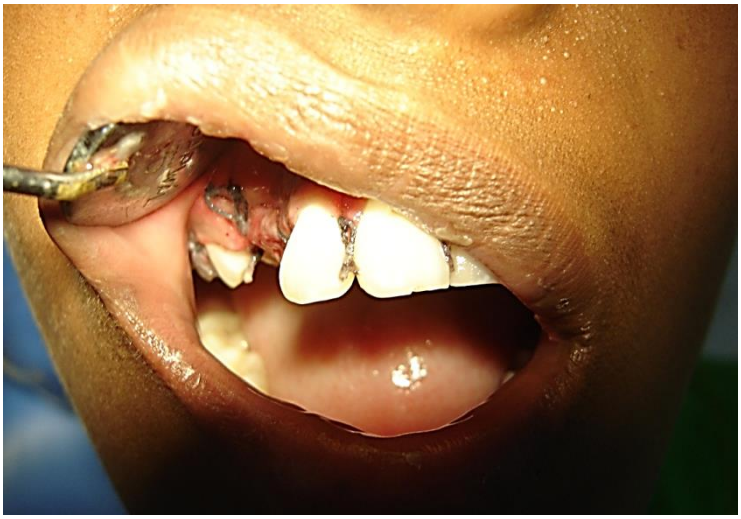


FIGURE 8: POST OPERATIVE DAY 1

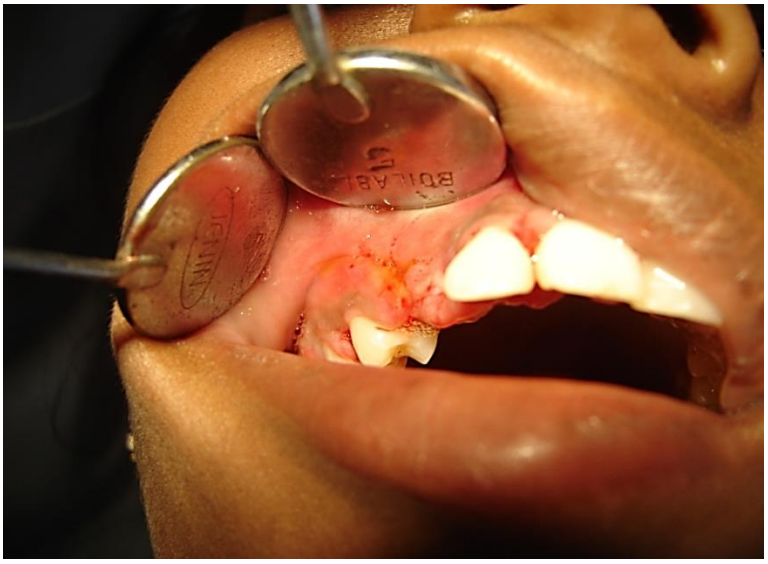


FIGURE 9: POST OPERATIVE 7 FOLLOWING SUTURE REMOVAL

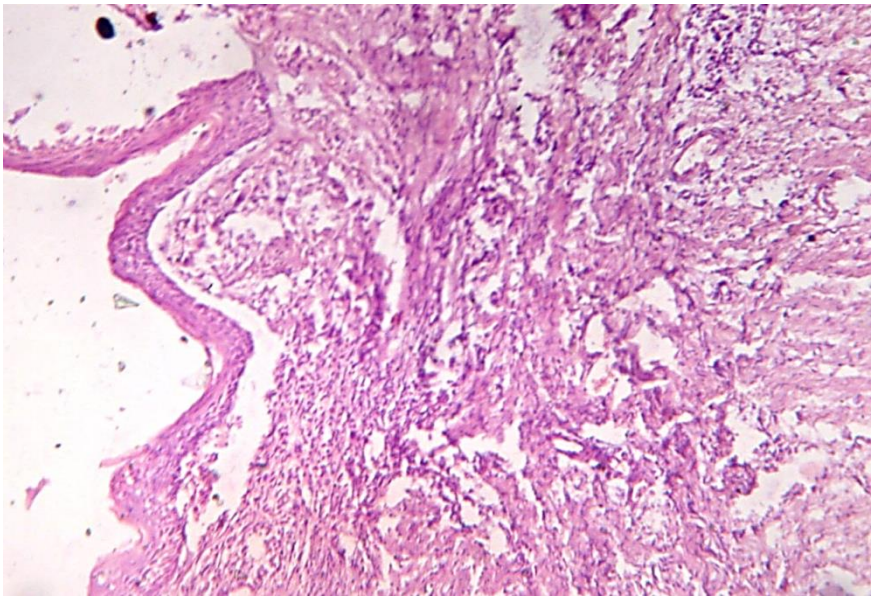


FIGURE 10: HISTOPATHOLOGICAL EXAMINATION OF THE LESION CONFIRMING DIAGNOSIS OF AOT

S.NO	AUTHOR	LOCATION	SIZE	ASPIRATION	TREATMENT	INFERENCE
1.	Patel G B V Et Al in 2022;	Left anterior maxilla	2cm *3cm	Positive	Surgical Excision	AOT is rare proper diagnosis is essential for treatment
2.	Sharma N et Al in 2012	Right anterior mandible	6cm*5cm	Positive	Surgical Excision	Incidence of AOT in mandible is less it must be considered as a differential diagnosis for odontogenic
3.	Nguyen Quang Duc et Al in 2022	Right Anterior Mandible	22cm *25 cm	Not done	Surgical Excision followed by Reconstruction with Osseocutaneous Flap	Size of the tumor was extensive and it was a rare case making reconstruction difficult with functional defect present post reconstruction.
4.	Dhupar V et Al in 2016	Left Maxilla	7cm*6cm	Not done	Subtotal Maxillectomy followed by reconstruction with tempoalis myofascial flap	Atypical features was seen along with its aggressive behavior therefore aggressive treatment had to be undertaken
5.	Dr Sushma Mehkri et Al in 2010	Right and left Maxilla	3cm*2cm	Not done	Incsional biopsy was done followed by enucleation of the tumor along with corresponding deciduous tooth and permanent tooth buds	Large bilateral presentation is a rare presentation of this type of tumor.
6.	Dr. Sonu Nigam et Al in 2005	Left Maxilla	4cm*4cm	Positive	Excisional Biopsy	Important to consider AOT as a differential diagnosis
7.	Mutalik VS et Al 2012	Left anterior maxilla	2.0 cm × 2.5 cm	Positive	Enucleation	Histopathology revealed nests and rosettes cell patterns. Solid areas, duct-like pattern, whorled arrangement of cells, and tubular appearance was evident
8.	Simarpreet V Sandhu et Al 2010	Right Maxilla	6 cm × 5 cm	Positive	Enucleation	AOT of maxillary antrum is extremely rare, a case of AOT that originated in the wall of a dentigerous cyst of the maxillary antrum,
9.	Agnes Assao et Al 2017	Right Maxilla	3 cm*5 cm	Positive	Enucleation	AOT of Maxillary antrum that resembled an odontoma with aggressive behavior

CONCLUSION

AOT a hamartomous odontogenic tumor with cystic characteristics usually remains undiagnosed unless swelling is markedly pronounced. The intraosseous slow growth of the tumor leads to bone resorption and teeth displacement requiring extraction. A need for sequential clinical and radiological examination and correlation will eliminate misdiagnosis and facilitate early intervention.

REFERENCES

1. Steensland HS. Epithelioma adamantinum. J Exp Med 1905;6 (4-6):377–389. DOI: 10.1084/jem.6.4-6.377
2. Kalia V, Kalra G, Kaushal N, et al. Maxillary adenomatoid odontogenic tumor associated with a premolar. Ann Maxillofac Surg 2015;5(1):119–122. DOI: 10.4103/2231-0746.161118
3. 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

4. Philipsen HP, Reichart PA. Adenomatoid ododntogenictumour: Facts and figures Eur J Cancer B Oral Oncol. 1998;35:125–31
5. H.P. Philipsen, P.A. Reichart, Adenomatoid odontogenic tumour: facts and figures, Oral Oncol. 35 (2) (1999 Mar) 125–131.
6. Marx RE, Stern D. Odontogenic and Nonodontogenic Cysts. In: Marx RE, Stern D.Oral and maxillofacial pathology: a rationale for diagnosis and treatment.2nd ed. Chicago:Quintessence. 2012; 654-57.