Adenomatoid Odontogenic Tumor: Rare Clinical Presentation In A 20 Year Old Male

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ABSTRACT

Adenomatoid odontogenic tumours (AOTs) are rare odontogenic tumours that typically occur in the anterior maxilla and are painless. They are typically linked to unerupted teeth or dentigerous cysts. AOTs make about 3% of all odontogenic tumours.[1] It is distinct from other odontogenic tumours, with over two-thirds (69%) being diagnosed between the ages of 10 and 19; over half (53%) occurring in teenagers; and 21% occurring between the ages of 20 and 29. Women have been diagnosed with the tumour about twice as often as men. [2-3] The tumour is typically linked to teeth that have not yet erupted, usually lateral incisors or canines.[4]In terms of its etiology, the lesion starts from the odontogenic epithelium (remaining dental lamina or enamel organ), which in turn influences the odontogenic ectomesenchyme and produces dentinoid material.[5] It frequently results in shifting of neighbouring teeth and the expanding of surrounding bone. However, because the lesion grows slowly, patients may put up with the swelling for years before it manifests as a noticeable deformity.[6] The tumour typically shows up on radiography as a well-defined, unilocular radiolucency that may be connected to an unerupted tooth, generally a canine.[7]The preferred course of treatment has been thought to be conservative surgical enucleation. Following the full excision of the tumour, membrane-guided tissue regeneration is advised for periodontal intrabony abnormalities brought on by AOT. [8] AOT recurrence is quite uncommon. [9] The prognosis is therefore very good. Here, we describe a rare instance of AOT in a 23-year-old man.

INTRODUCTION

CASE PRESENTATION

A 23-year-old male patient reported to the OPD of department of Oral and Maxillofacial Surgery, Bharati Vidyapeeth Dental College and Hospital, Sangli of swelling on left upper gum region since last 1 year which suddenly grew in size in last 15 days.

No relevant findings were noted extraorally. Intraoral examination revealed solitary swelling about 2 x 2 cm (Fig 2), oval in shape was seen over the anterior maxillary attached gingiva region extending from free marginal gingiva of 22, 23 and 24 superiorly till 2.5 cm and mesiodistally from distal of 21 till mesial of 24, oval in shape with diffused margins. Overlying mucosa was reddish-pink in color with firm to hard consistency on palpation, without any tenderness. Mesial tilting of crown 23 and distal tipping of 22 was noted. Patient also reported of intermittent pain.

Blood investigations performed prior to planning of treatment, revealed no significant finding. Bleeding and clotting time were within normal limit, also the blood sugar levels were in the normal range. HIV and hepatitis B tests gave negative finding.

Radiographic examination revealed a well-defined intrabony radiolucency with sclerotic margins, causing displacement of associated lateral incisor and canine was seen in OPG (Fig 3).

A CBCT of the region was also done which revealed a well-defined radiolucency along with thinned cortical borders S/O Cystic Lesion. (Fig 4)

FNAC was done which revealed a dark straw-colored fluid indicating an infected cyst.

After thorough clinical and radiographic analysis following differential diagnosis were given:

- Globulomaxilary cyst:
- Lateral periodontal cyst:
- Calcifying odontogenic cyst:
- Calcifying epithelial odontogenic tumor:

TREATMENT

Surgical enucleation and curettage of the lesion was planned under general anesthesia taking into account the size of the lesion and its close proximity to the nasal mucosa. Endodontic treatment of canine and 1st premolar was performed prior enucleation. Enucleation was performed (Fig 5) and the encapsulated specimen was sent for histopathological examination. Primary closure was performed and postoperative antibiotic and analgesic was prescribed. Immediate Post operative OPG was also taken (Fig 5).

Subsequent histopathological report revealed it to be a Adenomatoid Odontogenic Tumor. Histological sections studied show an encapsulated tumor (Fig 6) having variably sized multiple nodules composed of spindled epithelial cells with minimal stroma with small amount of eosinophilic matrix depositions. Within these nodules are seen variably sized duct-like spaces lined by cuboidal to columnar epithelium with nuclei away from luminal surface (Fig 7). Periphery of tumor shows anastomosing cords of tumor cell. Adjacent unremarkable bone is also included in the sections studied. There is no evidence of malignancy in the sections studied.

OUTCOME AND FOLLOW-UP

Regular follow-up of the patient was performed and 4 months postoperative radiograph revealed satisfactory bone healing of the defect with no signs recurrence (Fig).

DISCUSSION

Steensland[10] originally reported AOT, a rather rare unique odontogenic tumour, in 1905. Nonetheless, this tumour has been referred to by a number of names. A list of all AOT nomenclatures used in the literature was created by Unal et al.[11] The disease now known as AOT has been referred to by a number of names in the past, including adenoameloblastoma, ameloblastic adenomatoid tumour, epithelioma adamantinum, and teratomatous odontoma. It is believed that the odontogenic epithelium is the source of the contentious AOT because of its preference for tooth-bearing bone. [2,-3]Only 3% of all odontogenic tumours are AOTs, which are slow-growing lesions that are more common in young girls in their second decade of life and have a 2:1 preference for the anterior maxilla over the mandible.[12] Although the lesions are usually asymptomatic, as in the case described here, they may result in cortical enlargement and displacement of the neighbouring teeth.

Intraosseous follicular, intraosseous extrafollicular, and peripheral are the three clinicopathological forms of AOT. The follicular type mimics a dentigerous or follicular cyst by exhibiting a distinct unilocular radiolucency connected to the crown and frequently a portion of a tooth's root that has not yet erupted. The well-defined, unilocular radiolucency is present between, above, or overlaid upon the roots of erupted, permanent teeth, as shown in the example above. The extrafollicular form is not linked to an unerupted tooth. Discrete calcific foci can be seen in radiographs of around two-thirds of intrabody variations of AOT, although in our case, there were no visible calcific deposits.[12]Interestingly, the histology of all AOT variations was identical; nevertheless, according to the WHO's histological type, AOT is classified as an odontogenic epithelial tumour with duct-like structures and variable degrees of inductive alteration in the connective tissue. [13]. The preferred course of treatment is conservative surgical enucleation.

Given that AOT is encapsulated, tumour enucleation yields good results with a nearly zero recurrence rate. The patient described had a great outcome and reacted nicely to the conservative

surgical approach. The AOT case study aligns with current literature and research, however it includes some unusual presentations, such as a male patient in his 20s who experiences intermittent pain when palpated.

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, Reichart PA. Adenomatoid odontogenic tumor: facts and figures. Oral Oncol 1998;35:125–31.
3. Reichart PA, Reichart P, Philipsen HP. Odontogenic tumors and allied lesions. 1st edn. London: Quintessence, 2004.
4. Dayi E, Gurbuz G, Bilge OM, et al. Adenomatoid odontogenic tumour (adenoameloblastoma). Case report and review of the literature. Aust Dent J 1997;42:315–18.
5. Neville BW, Damm DD, Allen CM, et al. Oral and maxillofacial pathology. 2nd edn. Philadelphia: WB Saunders, 2002:621–3.
6. Olgac V, Koseoglu BG, Kasapoglu C. Adenomatoid odontogenic tumor: a report of an unusual maxillary lesion. Quintessence Int 2003;34:686–8.
7. Ali K, Munir FM, Nazir A. Clinical presentation and management of adenomatoid odontogenic tumour. Pak Oral Dnt J 2006;26:163–5.
8. Blumenthal NM, Mostofi R. Repair of an intrabony defect from an adenomatoid odontogenic tumor. J Periodontol 2000;71:1637–40.
9. Philipsen HP, Reichart PA, Nikai H. The adenomatoid odontogenic tumor (AOT): an update. J Oral Med Pathol 1997;2:55–60.
10. Steensland HS. Epithelioma adamantinum. J Exper Med 1905;6:377–89.
11. Unal T, Cetingul E, Gunbay T. Peripheral adenomatoid odontogenic tumor: birth of a term. J Clin Pediatr Dent 1995;19:139–42.
12. John JB, John RR. Adenomatoid odontogenic tumor associated with dentigerous cyst in posterior maxilla: a case report and review of literature. J Oral Maxillofac Pathol 2010;14:59–62.
13. Kramer IRH, Pindborg JJ, Shear M. WHO histological typing of odontogenic tumors. 2nd edn. New York, Berlin, Heidelberg: Springer-Verlag, 1992.

PICTURES

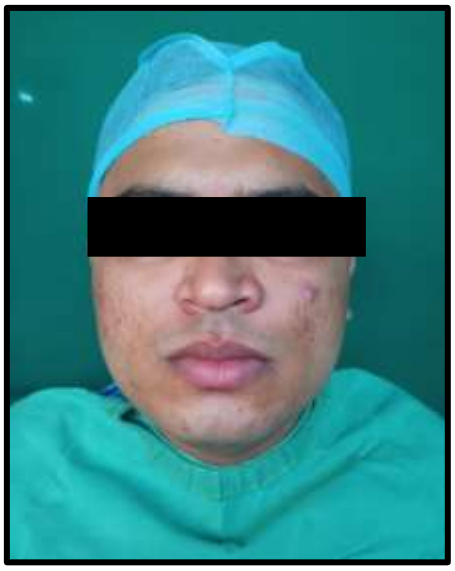


Fig 1: Extraoral picture

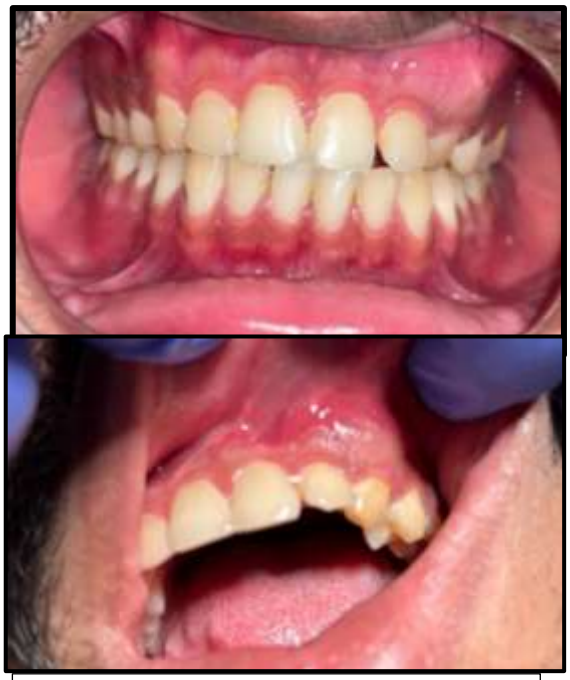


Fig 2: Intraoral picture showing the size and extent of the lesion

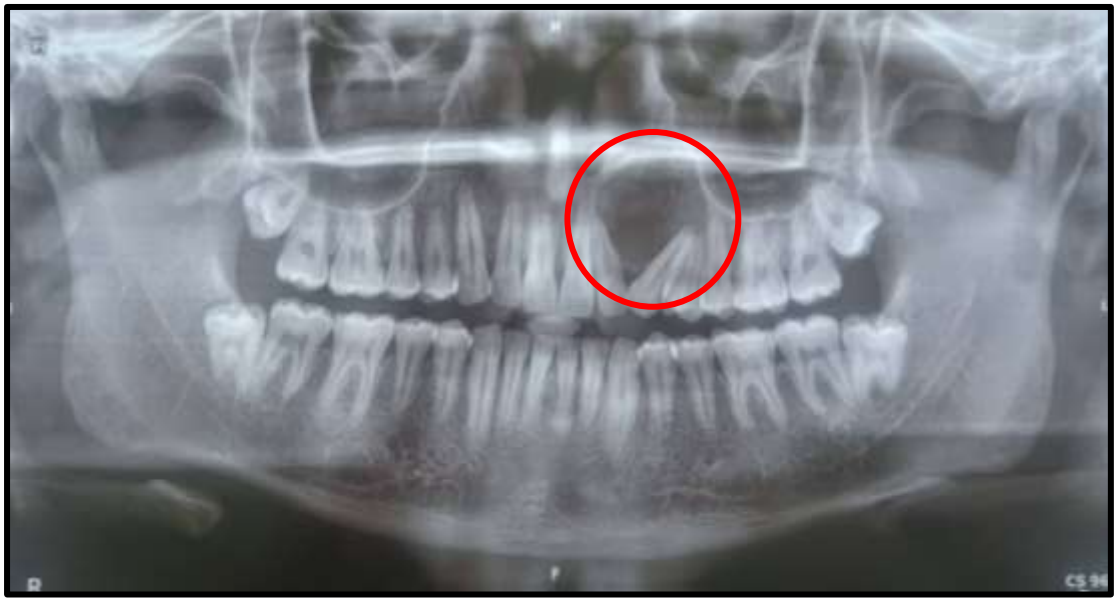
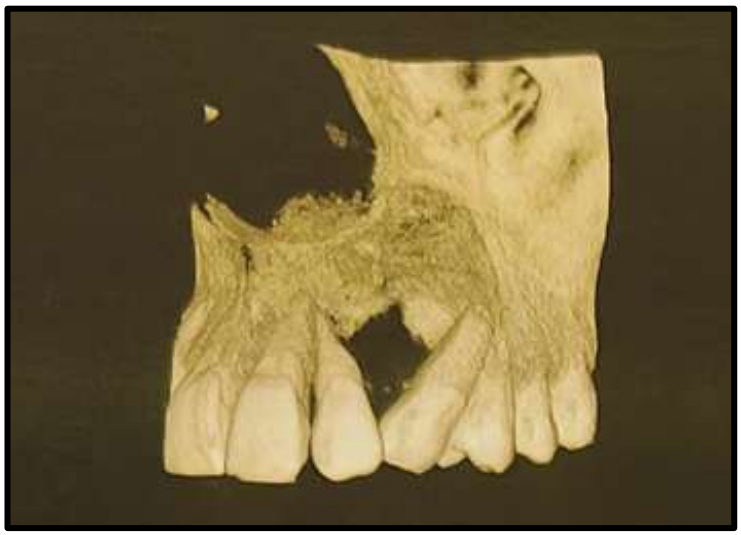
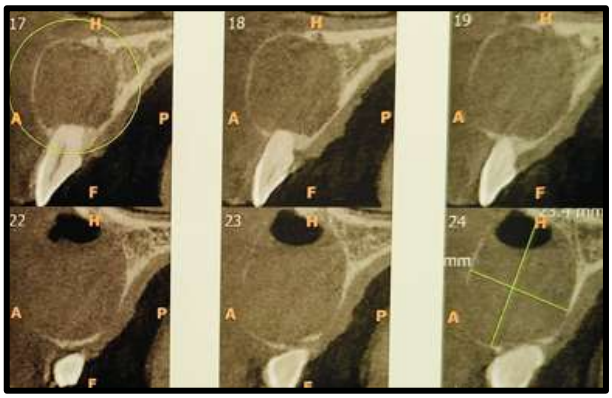


Fig 3: OPG showing well defined radiolucent lesion in between 22 and 23 with well-defined sclerotic borders





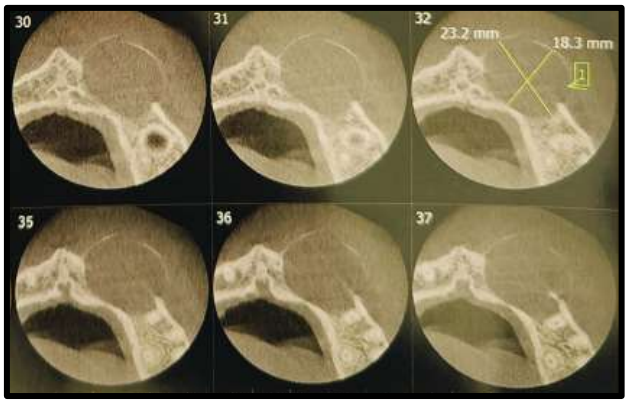


Fig 4: CBCT showing thinning of the labial cortical plate with dimensions of the lesion: 23.2 mm x 18.3 mm

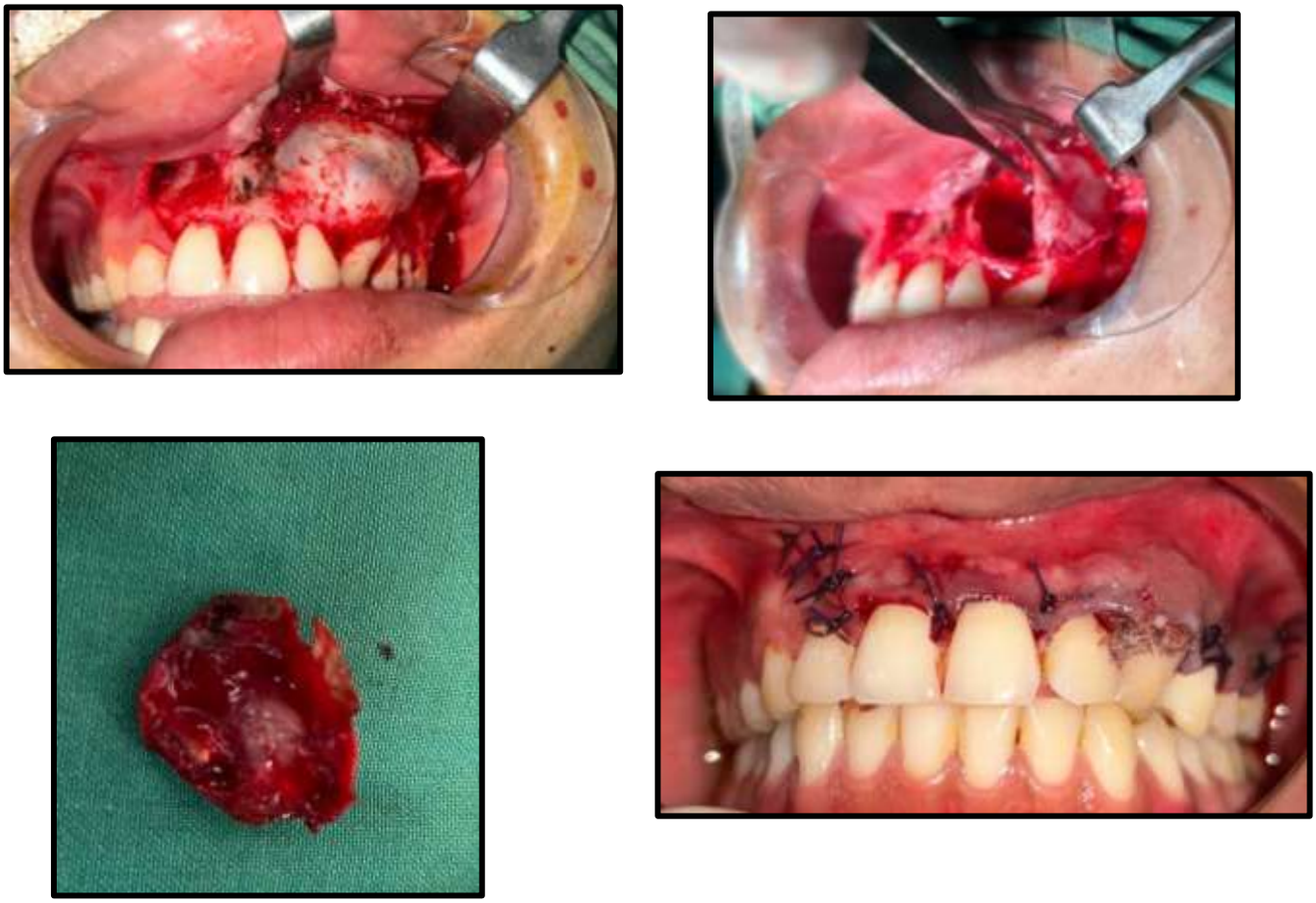


Fig 5: Intraoperative pictures showing the excision of the lesion

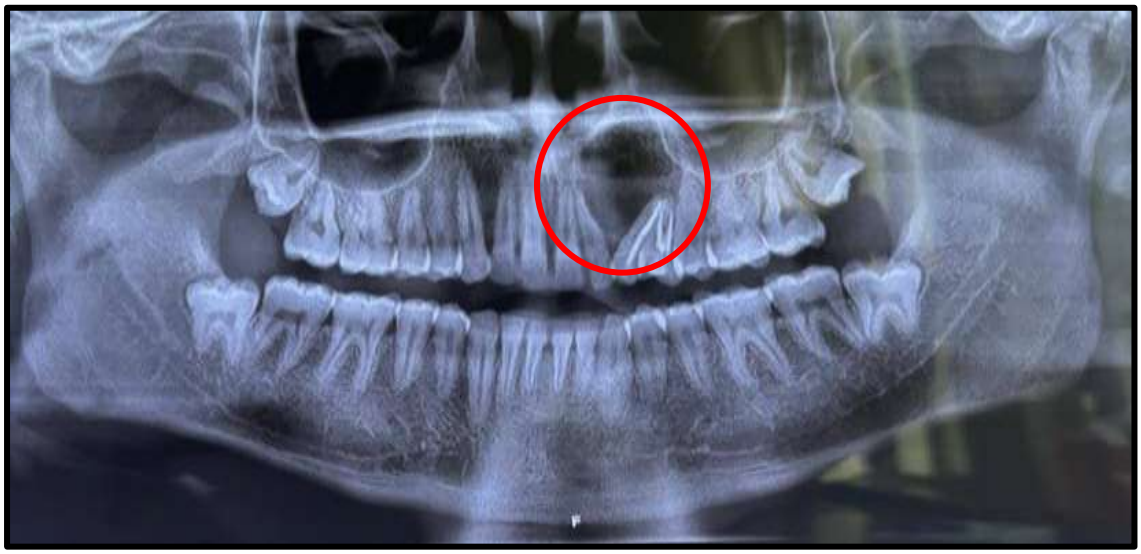


Fig 6: Immediate Postoperative OPG

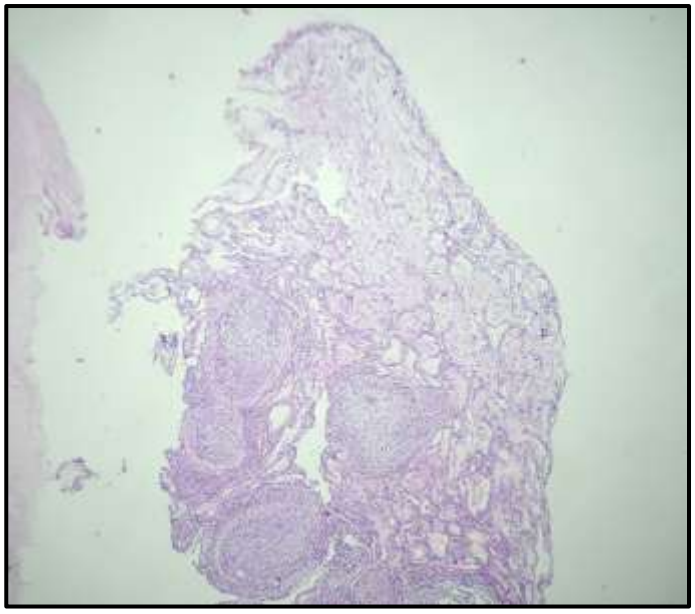


Fig 6: Low power image of encapsulated adenomatoid odontogenic (H&E, scanner view).

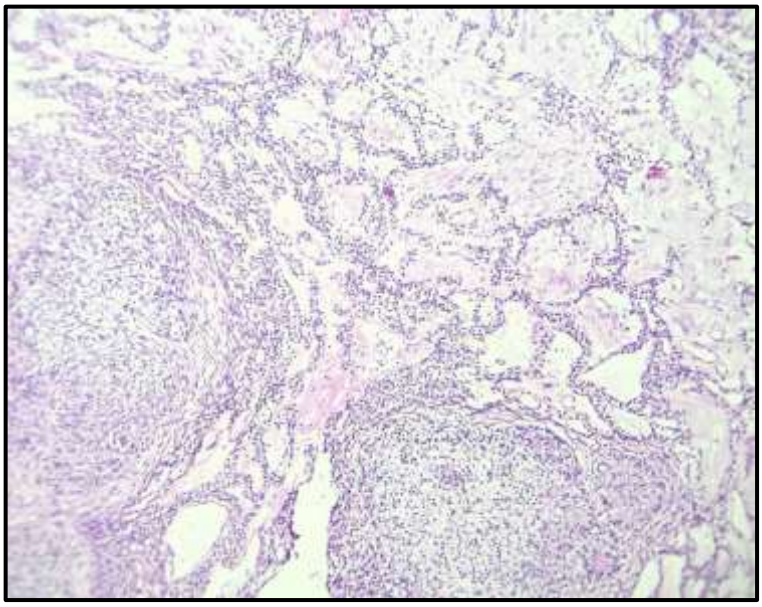


Fig 6: Nodular islands with a cribriform to nodular architecture. (H&E, 10x).

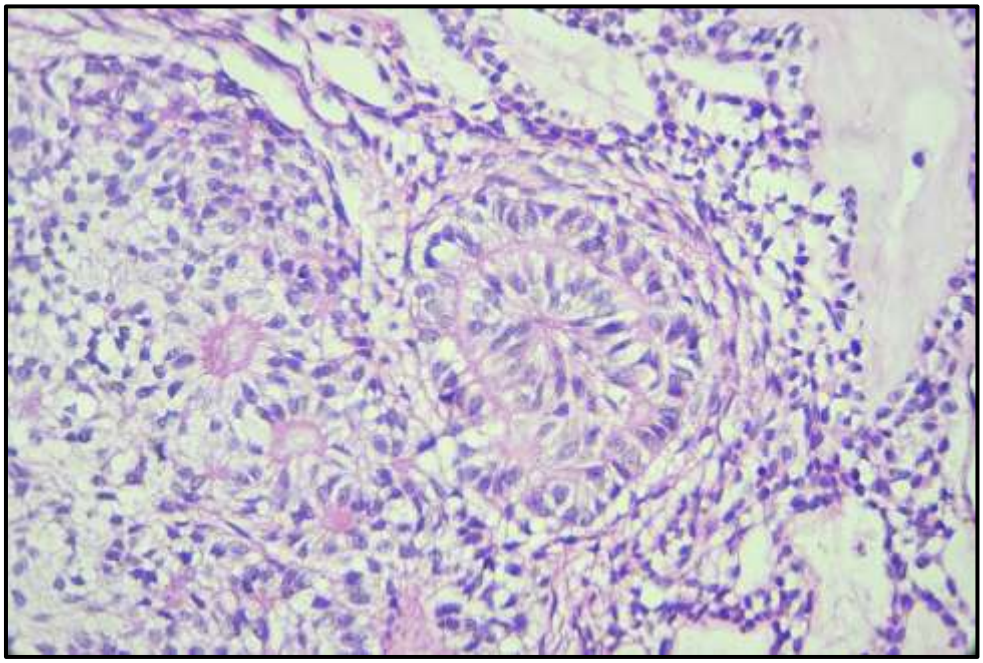


Fig 7: AOT with heterogeneous appearance with central rosette-like structure. H&E, 40x).