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RESEARCH ARTICLE

AN UNUSUAL CASE OF EXTRA FOLLICULAR ADENOMATOID ODONTOGENIC TUMOR: A CASE REPORT.

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Abstract

Adenomatoid odontogenic tumor (AOT) is an uncommon tumor of odontogenic origin. It has two clinicopathologic variants, namely intraosseous (follicular and extrafollicular) and extraosseous(peripheral). The extrafollicular variety is rare and shows a prevalence of 26.9%. It generally occurs in younger females with a mean age of 24 years in the anterior maxilla. A case of extrafollicular AOT with a few unusual features is being reported here. In the reported case, it occurred in an older female, 55 years of age in the anterior mandible. Conventional and advanced imaging revealed a well-defined mixed radiolucent -radiopaque lesion. Surgical enucleation was performed and the histopathological diagnosis was suggestive of extrafollicular AOT. The patient recovered well after the surgical procedure and has been on regular follow up.

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Case Report:-

A 55-year-old female patient reported with the complaint of pain in the lower right front teeth region since 1 week. The pain was primary in incidence, gradual in onset, initiated on chewing, moderate in intensity, intermittent and localized. She had temporary relief from pain on consuming analgesics. Medical history revealed that the patient was diabetic since 7 years and was on oral hypoglycemics. On general examination, the vital signs were within normal range. No abnormality was detected on extra-oral examination. On intra oral examination of hard tissue, deep cervical abrasion was present in relation to 44,45 (Fig1). Moderate attrition was noted in relation to 43,45. No mobility of teeth were noted. Tenderness on vertical percussion was present in relation to 43,44,45. On soft tissue examination, vestibule in relation to 43, 44, 45 was obliterated, bony hard and tender on palpation. No bleeding or

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pus discharge were observed on digital pressure.

A provisional diagnosis of infected radicular cyst was given. Calcifying odontogenic cyst, Traumatic bone cyst, Ossifying fibroma and Calcifying epithelial odontogenic tumor were considered in the differential diagnosis as these lesions commonly occur in females, in the mandibular premolar region and present as a bony hard swelling.

On electric pulp testing, 43 showed delayed response and 44,45 responded normally.

Orthopantomograph revealed a circular, well-defined, mixed radiolucent radio-opaque lesion in the inter-radicular region of 43 and 44 approximately measuring 2x2 cm (Fig3). Superio-inferiorly, it extended 3mm beneath the alveolar crest upto the apex of 43 and 44. Medio-laterally, it extended from distal aspect of root of 44 to mesial aspect of root of 43. Internal structure revealed specks of calcification. Displacement of roots of 43 and 44 were noted.

CBCT revealed a well-defined radiolucent lesion with clearly defined radiopaque margins in the inter-radicular region of 43 and 44 measuring approximately 11.54mm x 12.40mm x 13.52mm (Fig4a). Expansion of buccal and lingual cortical bone plates was noted (Fig4b). Root divergence of 43 and 44 was noted with no root resorption. Radiographic DD of Calcifying cystic odontogenic tumor, Adenomatoid odontogenic tumour, Calcifying epithelial odontogenic tumour and Ossifying fibroma were considered. However, root resorption is common in CCOT, CEOT and ossifying fibroma and appearance of calcifications varies in each.

Following the investigations, extraction of 43 and 44 was done. Enucleation of the lesion was performed under local anesthesia (fig5). Suturing was done and primary hemostasis was achieved.

On macroscopic examination, the tissue specimen was well circumscribed and spherical in shape measuring about 1.3cm attached to a small bony spicule. The color appeared pinkish-grey. The mass was soft to fluctuant in consistency.

The Hematoxylin and eosin stained sections revealed a thick fibrous capsule, a nodular proliferation of cuboidal, columnar, spindle and polygonal cells. The cells were arranged in nodules, strands and plexiform patterns. Eosinophilic droplets were seen between these cells. Cystic spaces lined by flattened cells, few duct-like structures and few rosettes were also observed. Eosinophilic material and calcifications of varying sizes were present. Areas resembling calcifying epithelial odontogenic tumour (containing polyhedral squamous cells with prominent intercellular bridges) were also seen.

Discussion:-

The occurrence of extra-follicular type is quite rare with a prevalence of 26.9% compared to follicular type which accounts for 70.8% of all AOTs². The pathogenesis of AOT is not clear and a number of hypotheses have been proposed. Theoretically, it is said to arise from the enamel organ, the epithelial lining of the dentigerous cyst, epithelial rests of Malassez of the deciduous or permanent tooth, or remnants of the dental lamina and may show an ameloblastic phenotype⁴. However, Philipsen et al also argued that all AOT variants show identical histological features and therefore it points towards a common origin from the dental lamina or its remains.⁵

A Pubmed search of extra-follicular AOT revealed 35 case reports of extra-follicular AOT till date out of which detailed case history was available only in 13 cases (13+22). (Included in Table 1.)

Of these 13 case reports, 8 cases were seen in women indicating a slight female predilection. It was mostly reported in young adolescent, however, 4 cases were seen in middle aged. The number of cases occurring in maxilla was 7 and the rest in mandible suggesting no definite preference of maxilla or mandible. Out of 13 cases, 7 cases were seen in the anterior region and aggressive forms were seen in 4 cases. Most of them presented as a painless hard swelling of the involved jaw bone. From the above findings, we can infer that there has been a change in the trends with regard to clinical presentation in AOT and variations have been observed from what has been believed conventionally. The present case also seemed to show a few variations from previous literature as it occurred in an older female (55 years) in the anterior mandible and was associated with pain.

Radiographically, extrafollicular AOTs can be divided into 4 subdivisions based on its location and relationship with

adjacent teeth viz., E1 - No relation to tooth structure either erupted or unerupted, E2- Intra-radicular adjacent roots diverge apically due to tumor expansion, E3- Superimposed on the root apex and E4- superimposed on the mid-root level. All 13 cases presented as circumscribed radiolucent area with a well- defined corticated or sclerotic border. Internal structure showed fine calcifications, flocculent pattern and patchy calcifications in 3 cases. Expansion of cortical plate was seen in 4 cases. The size of the intraosseous lesions varied from 0.8cm to 4cm. Root resorption was seen in 6 cases and root deviation in all the cases. Majority of them belonged to E2 variant and 1 case belonged to E3 variant. The present case belonged to the E2 variant with typical radiographic features.

Histologically, all the variants of AOT are seen to have the same histological features. It may present as a solid mass or as cystic spaces which is well- encapsulated. Duct-like structures are seen lined by a single row of columnar epithelial cells giving a rosette-like pattern. Spindle-shaped or polygonal cells are seen forming sheets and whorled masses in a scant connective tissue stroma. The lumen may be empty or filled with amorphous eosinophilic material. Dystrophic calcification is usually encountered in most cases.

In the present case, along with the above described histopathological features typical of AOT (fig6a), a few areas of the tissue sample contained polyhedral squamous cells mimicking a CEOT (fig6b). Review of literature supports the presence of CEOT like feature in few cases of AOT.¹⁹

In the last few years, several studies have reported immunohistologic properties of AOT. The classical AOT phenotype is characterized by a cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium based on positive staining with CK5, CK17, and CK19. Recently, Crivelini, *et al.* detected the expression of CK14 in AOT and concluded that this probably indicate its origin in the reduced dental epithelium.⁸

The surgical management of this tumor is usually enucleation along with the associated impacted tooth and simple curettage. Conservative treatment is adequate because the tumor is not locally invasive, is well encapsulated, and is separated easily from the bone. The recurrence rate is as low as 0.2%⁹. In addition, the use of lyophilized bone and guided tissue regeneration are recommended in large osseous cavities⁷. The prognosis is usually excellent in majority of the cases.

Conclusion:-

It may be inferred from the present case that AOT does not always occur in a narrow age range and in restrictive sites and can show variations in its presentations. Hence, it is suggested that AOTs must be considered in differential diagnosis of mixed radiolucent radio-opaque lesions occurring in older age group, anterior mandible, the absence of an impacted tooth.

Table1:- Clinical and Radiographic Features of extra-follicular AOT as available in PubMed Search:

	YEAR	CLINICAL FEATURES	RADIOGRAPHIC FEATURES
1	1996	35/Male. Painless intra-oral swelling extending from 35 to 45. Bucco- lingual expansion. 31,32,41,42 were mobile. Reduced response of the involved teeth to cold in vitality tests. ¹⁸	A well-demarcated radiolucency with scalloped margins. Patchy areas of calcification in internal structure . 35 to 45 showed root resorption
2	2002	15/Female. No extra oral/intra oral swelling. Related 11, 21 were vital. ¹¹	Inter-radicular region of 11,21. Well-defined, round to oval radiolucency(1.5 x1 cm) with a radiopaque margin . No deviation of root.
3	2007	22/Female. Painless and circumscribed swelling of the right maxillary region . ¹⁵	Periapical region of 12 to 17. Unilocular well-defined radiolucent lesion at the apices of the roots from 12 to 17. Root resorptions from 12 to 17.
4	2007	40/Male. A bony hard swelling from 36 to 45 with eggshell crackling. Buccal and lingual cortical expansion. ¹⁷	A well-defined radiolucent lesion extending from tooth 36 to 45 with root resorption of the incisor teeth. (Origin from unicystic ameloblastoma)
5	2009	13/ Female. No extra oral/intra oral swelling. Related 43 and 44 were vital. ⁶	Inter-radicular region of 43 ,44. well-defined, unilocular radiolucency (0.8X1 cm) with

			slightly radiopaque margins with displacement of the roots . No evidence of internal calcifications. Deviation of roots of 43 and 44.
6	2009	15/Female. No extra oral deformity. Painless intra oral swelling in 32-41region with mild labio-labial expansion. 32 was non-vital. ¹⁰	inter-radicular region of 32,41. Circumscribed unilocular radiolucent area with fine calcifications. Labiolingual expansion with thinned cortical layer. Root deviation of 32 and 41
7	2011	34/Male. Extra oral swelling. Intra-oral swelling of 21,22,23 involving both both labial and palatal mucosa with vestibular obliteration. 22 and 23 were slightly displaced. ¹²	Inter-radicular region of 22,23. Well- defined unilocular radiolucency with thin sclerotic border . Displacement of 22, 23. Root resorption of 23,24
8	2012	17/Female. Extra oral swelling. Painless bony hard swelling in 41 to 46. 42,43,44 and 45 were mobile and non vital. ⁸	41 to 46 region. Well-circumscribed radiolucency . Deviation of roots of 43,44 and root resorption of 44,45,46
9	2013	19/Female. Solitary intra-oral swelling over the gingiva of 21 and 22 . Displacement of 21,22. Bony hard painless swelling. ¹⁴	Inter-radicular region of 11,12. well-defined radiolucency with sclerotic margins. Occlusal radiograph revealed expansion of labial cortex. Deviation of root of 11,12
10	2013	18/Female. intra oral swelling in 12, 13 region. Bony hard painless swelling. ¹³	Inter-radicular region of 12, 13. well-circumscribed unilocular radiolucency(1.4×1.0 cm). Increase in the interdental space of 12,13. Deviation of root of 12,13.
11	2013	18/Female. Extra oral swelling. Intra oral painless bony hard swelling in the left mandibular ramus and retromolar area with obliteration of buccal vestibule. 37 was mobile. ⁹	Inter-radicular region of 44 to 47 region. Unilocular, oval, well defined radiolucency of 4x5cm with well defined sclerotic borders. No evidence of internal calcification.Deviation of root of 47
12	2015	24/Male. Asymptomatic bony hard expansion in relation to the buccal aspect of 13 and 14. ¹⁶	Oval well-defined unilocular radiolucency (2 x 1.7 cm). target-like appearance with a central area of floccular calcification. Divergence of the roots of the 13, 14 with no root resorption.
13	2016	50/Male. Extra oral swelling. Painless swelling on right side of hard palate from 11 to 16 region. ⁷	11 to 16 region. A well-defined unilocular radiolucency with sclerotic border(4x3 cm). Buccal cortical expansion and multiple foci of calcifications. Displacement of 12, 13 and root resorption of 11, 21. Obliteration of maxillary sinus.
14	2017 (Present case)	55/Female, Intra oral buccal vestibule obliteration from 43,44 to 45 associated with pain. Associated teeth was vital.	Inter-radicular region of 43,44. well-defined radiolucency with radio-opaque border (2X2cm). Internal specks of calcifications. Buccal and lingual cortical plate expansion.Root deviation of 43, 44



Fig1:- intra- oral picture of mandibular right quadrant.

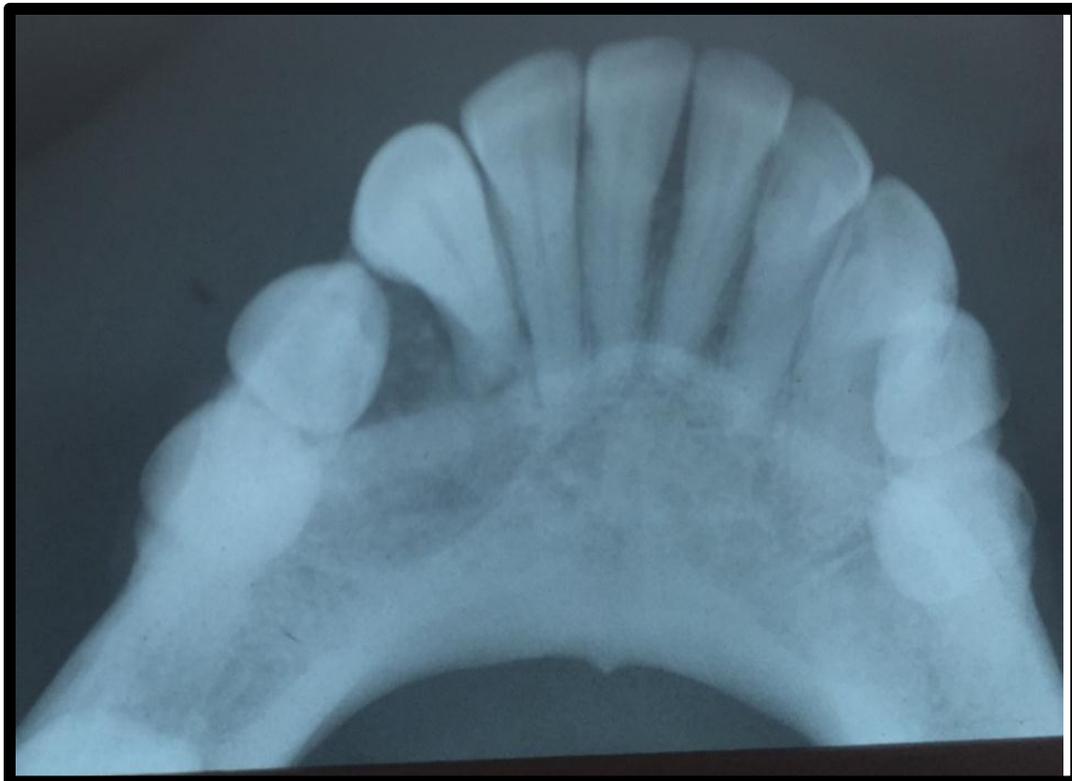


Fig2:- Occlusal radiograph showing well defined radiolucency in the inter-radicular region of 43 and 44.



Fig 3:- OPG showing a well-defined, circular, mixed radiolucent radio-opaque lesion in the inter-radicular region of 43 and 44.

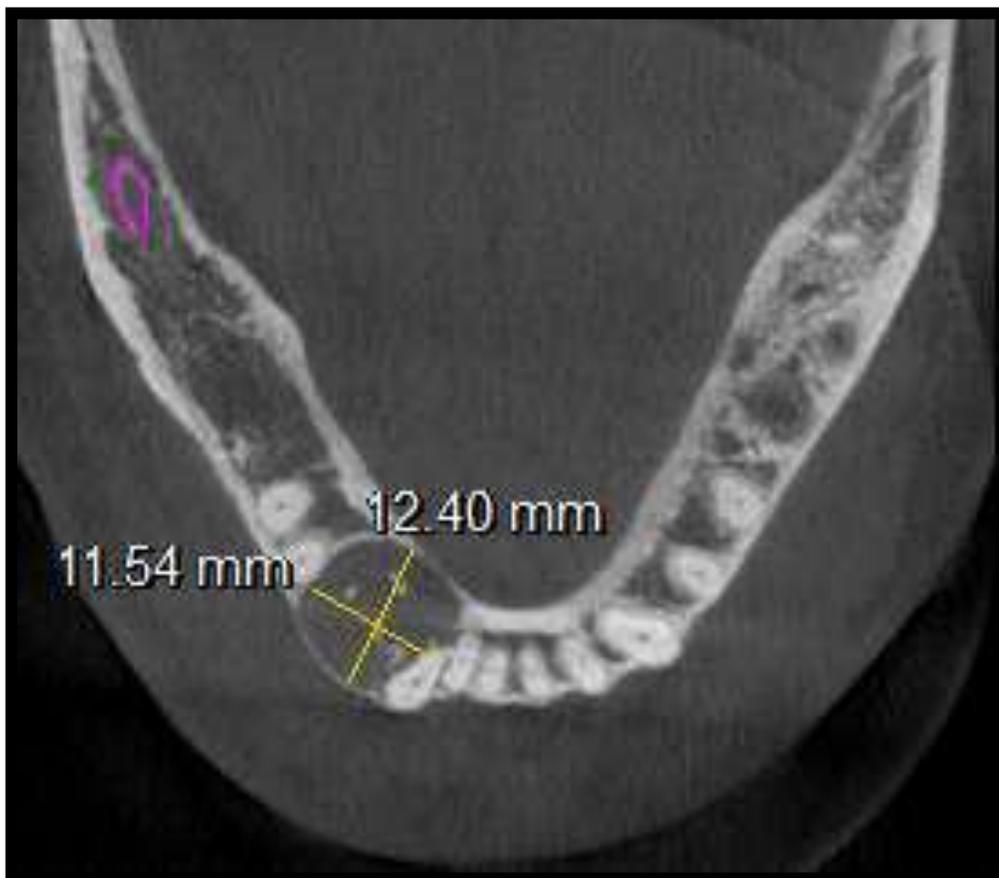


Fig 4a:- An axial image of CBCT displaying a round shaped well defined radiolucent lesion with clearly defined radiopaque margins and expansion of buccal and lingual cortical plates.

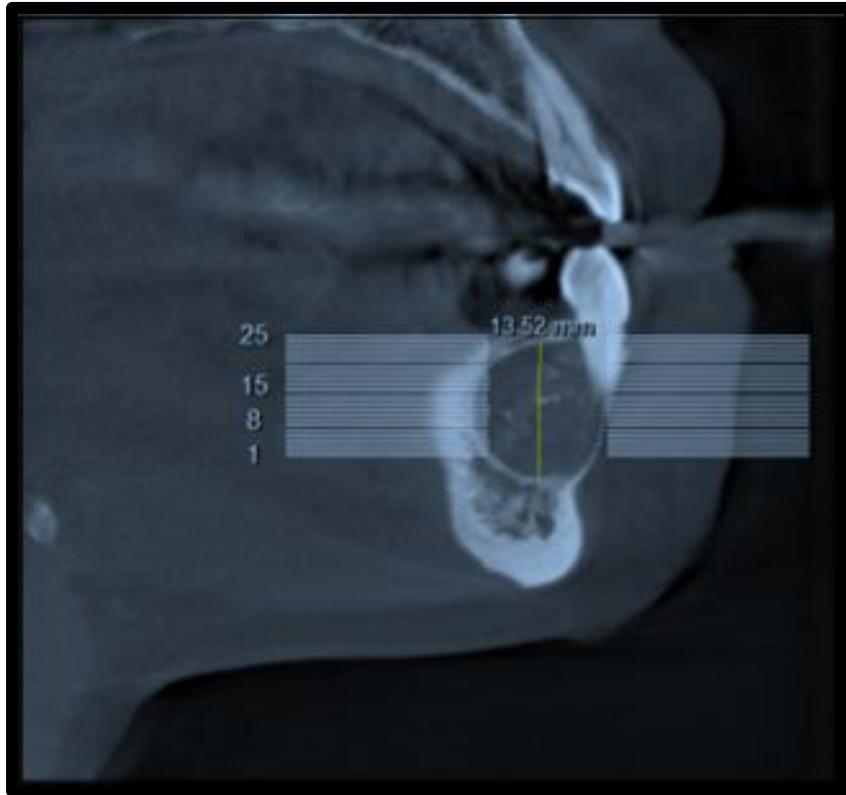


Fig 4b:- Sagittal view of the lesion in CBCT showing the superior-inferior extension of the lesion.

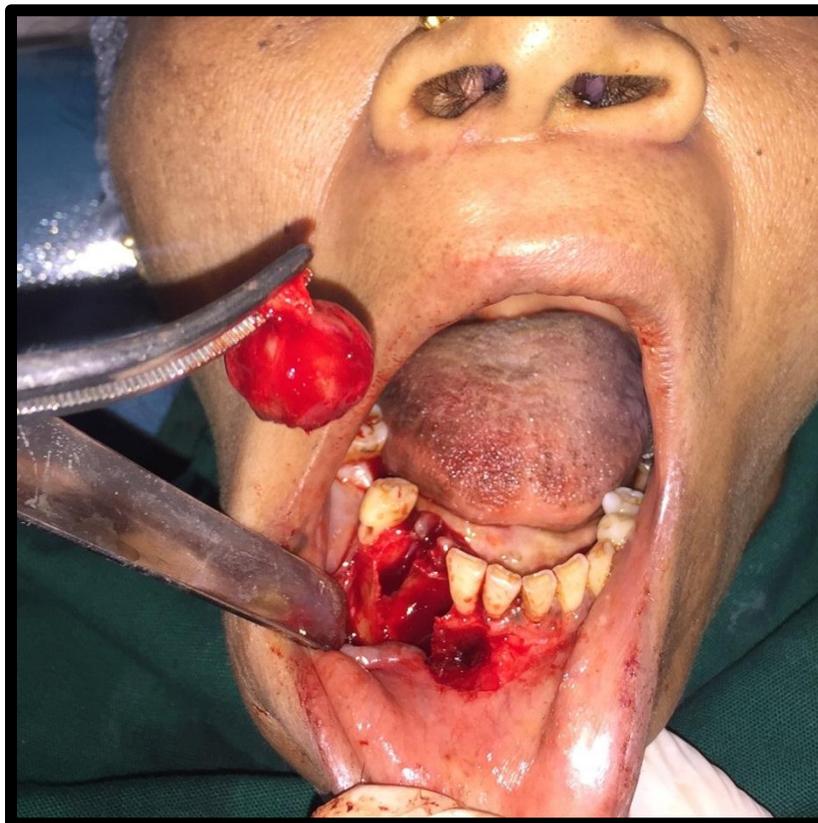


Fig 5:- The enucleated mass was spherical in shape measuring approximately 1.3 cm.

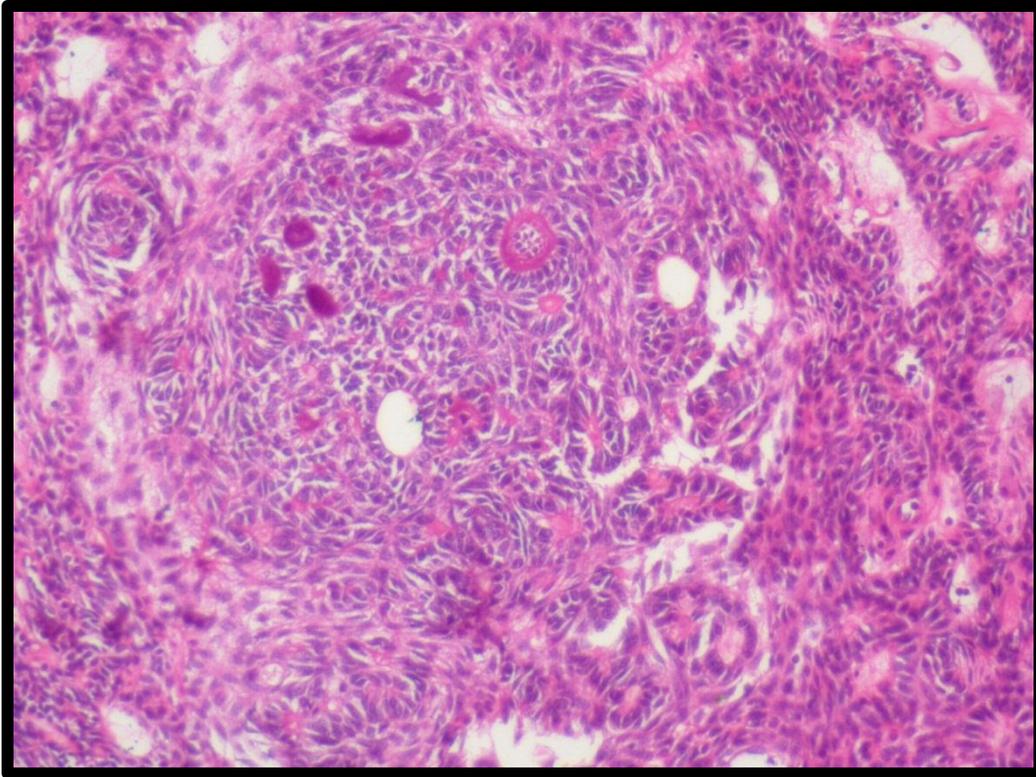


Fig6a:- Histopathological picture showing A: tumor droplets and areas of calcifications. B: Cystic space with rosette like pattern

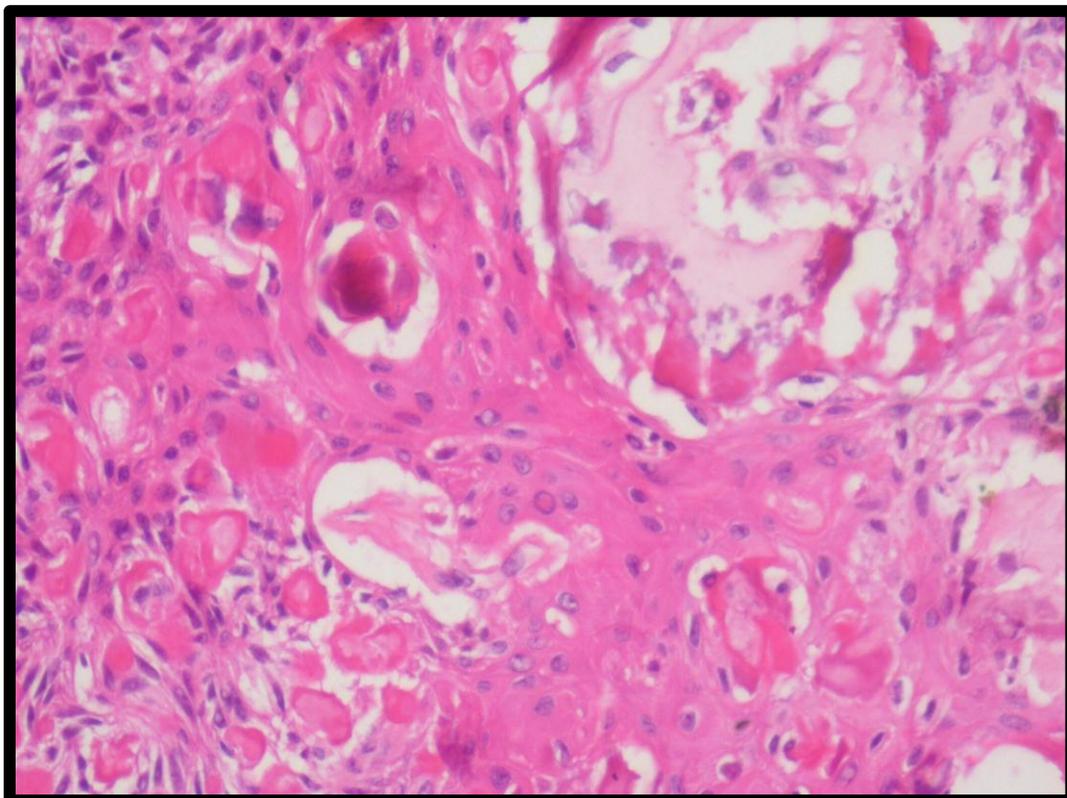


Fig6b:- Areas resembling calcifying epithelial odontogenic tumor containing polyhedral squamous cells.

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