# ADENOMATOID ODONTOGENIC TUMOUR: GROSS AND HISTOLOGICAL EXAMINATION OF 45 CASES

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## SYNOPSIS

**This paper represents a reappraisal of the gross and histological features of 45 cases of adenomatoid odontogenic tumours as observed under conventional light and fluorescence microscopy. The findings conformed largely to those of previous studies. Usage of the term adenomatoid odontogenic tumour in preference to its old name 'adenoameloblastoma' is emphasized. The differential diagnosis of this entity from the ameloblastoma and salivary gland tumours is discussed.**

## INTRODUCTION

The adenomatoid odontogenic tumour (AOT) is an uncommon odontogenic epithelial tumour with characteristic clinical and histological features. Although it was first recognised as a distinct clinicopathological entity by Stafne in 1948 (1), the original description of this lesion was by Ghosh in 1934 (2). Since then more than 160 verifiable cases have been reported worldwide (1-12), a 100 of these were described under different names including adenoameloblastoma (3), cystic complex composite odontoma (4), tumour of enamel organ epithelium (5), ameloblastic adenomatoid tumour (6) and adamantinohaemangioma (7).

Most reports have consistently shown that the AOT commonly presents as a slowly growing, painless mass often involving the anterior maxilla (8-12). It is found more frequently in female patients and has a peak incidence in the second decade of life (8-11). Radiographically, the tumour is commonly associated with an unerupted tooth, simulating a dentigerous cyst (8-12). However there is an increasing recognition that the calcification sometimes present in this tumour may produce a faintly detectable radiopacity which may be helpful in reaching a preoperative diagnosis (10).

The histological features of the AOT have been well described (1-12). The most frequent pattern is a proliferation of sheets, nests and cords of ameloblast-like cells supported by a scanty often haemorrhagic stroma. These cells may be organised to form whorls, rosettes, nodules or surround ovoid spaces to form duct-like structures. Additionally, "amyloid"-like eosinophilic material may be found filling or lining the ductal lumens or disposed as intercellular droplets in cellular areas. Calcific materials is often found throughout the tumour and appears to develop primarily at the junction between tumour epithelium and adjacent vascular stromal tissue.

While the clinical and histological features of the AOT are well-documented, considerable controversy exists as regards the origin of this lesion. Nevertheless the general consensus of opinion is that the AOT is a lesion of odontogenic epithelial origin. This is because the tumour is found exclusively in the jaws, is frequently found in association with dentigerous cysts or impacted teeth, and has cellular characteristics similar to those of the various components of the enamel organ (10).

Because the AOT is also widely known by its other name, adenoameloblastoma which is a misleading term, this tumour must be distinguished from the ameloblastoma. The latter has a more guarded prognosis, and a histological diagnosis of ameloblastoma may eventuate in disfiguring surgical procedures. The AOT on the other hand are benign harmatomatous lesions that can be adequately treated by simple surgical excision (10). It is the intent of this study to present the gross and microscopic findings observed in 45 cases of AOT so that the histological parameters of this entity can be more readily appraised.

## MATERIAL AND METHOD

Forty-five cases of AOT were obtained from the files of the Division of Stomatology, Institute for Medical Research, Kuala Lumpur. These represented lesions diagnosed in this department between 1968 and 1986. The criteria for the selection of these cases was based on the definition of an adenomatoid odontogenic tumour by the WHO committee for the Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions (13).

> "A tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumour may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst."

The cases in this study were from patients ranging in age from 4 to 31 years. There were 28 female and 15 male patients; in two cases the sex was unrecorded. These consisted of 23 Malays, 10 Chinese, 7 Indians, 1 Sikh, 1 Kadazan, 1 Bugis and 2 of unknown race. Thirty-three of these tumours were from the maxilla and 12 from the mandible.

Serial sections stained with haematoxylin and eosin, Periodic acid Schiff test and thioflavine T were analysed in most cases; in some cases only a few sections were available. The histological findings were made without reference to the official reports of these cases.

## RESULTS

### Macroscopic Findings

Forty-four of the 45 surgical specimens were soft tissue curettings or enucleated cystic sacs (Fig. 1); one case consisted of a resected portion of the body of the right mandible. Twenty-one of these soft tissue

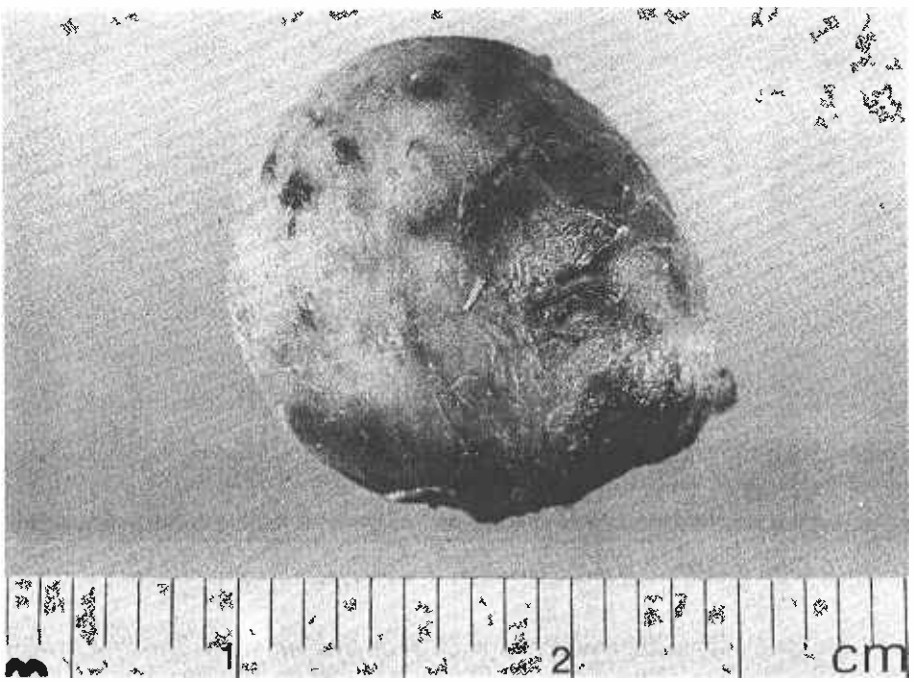


Fig. 1 Gross appearance of an enucleated adenomatoid odontogenic tumour

specimens were associated with an unerupted tooth (Fig. 2): 17 of these cases showed attachment of the cyst lining to the amelocemental junction of the tooth (this being a canine in 12 cases; lateral incisor in 2 cases; central lower incisor in 1 case; maxillary molar in 1 case; and lower second premolar in 1 case); 1 case was attached to the root of a lower canine; and in the remaining 3 cases the site of attachment was not specified.

Analysis of the cyst content revealed that most of these contained granular tissue (Fig. 3) while in 3 cases these were described as whitish, friable and solid.

In 39 soft tissue specimens the size of the lesion was specified. This may ranged from 1 cm to 5 cm with a mean of 3 cm and a median of 3.3 cm.

Microscopic Findings

Low power view examination revealed that the majority of these soft tissue specimens presented as discrete, well-encapsulated lesions (Fig. 4). The tumour masses may be focal in distribution (Fig. 5) or formed a mural lining backed by a fibrous connective tissue wall (Fig. 6). For those solid lesions, the tumour masses may entirely fill the lumen (Fig. 4), formed locules separated by intervening fibrous septa (Fig. 7) or occurred as luminal proliferations projecting into the cystic space (Fig. 8).

These tumour masses characteristically consisted of ameloblast-like cells commonly disposed to form sheets, nests and cords of epithelial cells (Fig. 9) supported by a scanty often haemorrhagic connective tissue stroma. Within these sheets of tumour cells, solid areas consisting of whorls and rosettes, and duck-like spaces may be identified (Fig. 10). These duct-like structures were characteristically lined by tall columnar cells exhibiting polarisation of their nuclei away from the lumens. The rosettes frequently showed a peripheral row of columnar or cuboidal cells with palisading of their nuclei (Figs. 10 and 11). Between opposing rows of cells within these rosettes and also lining the duct an amorphous, eosinophilic, PAS-positive material of varying quantity may be present (Figs. 10 and 11). This may also occur as intercellular droplets in the solid tumour areas (Fig. 12). In adition, foci of deeply basophilic, often spheroidal calcific material may be found scattered within the tumour masses (Fig. 13) or at the junction between tumour epithelium and adjacent stromal tissue (Fig. 10). The calcific foci fluoresced strongly in thioflavine T stained sections viewed with ultraviolet light (Fig. 14).

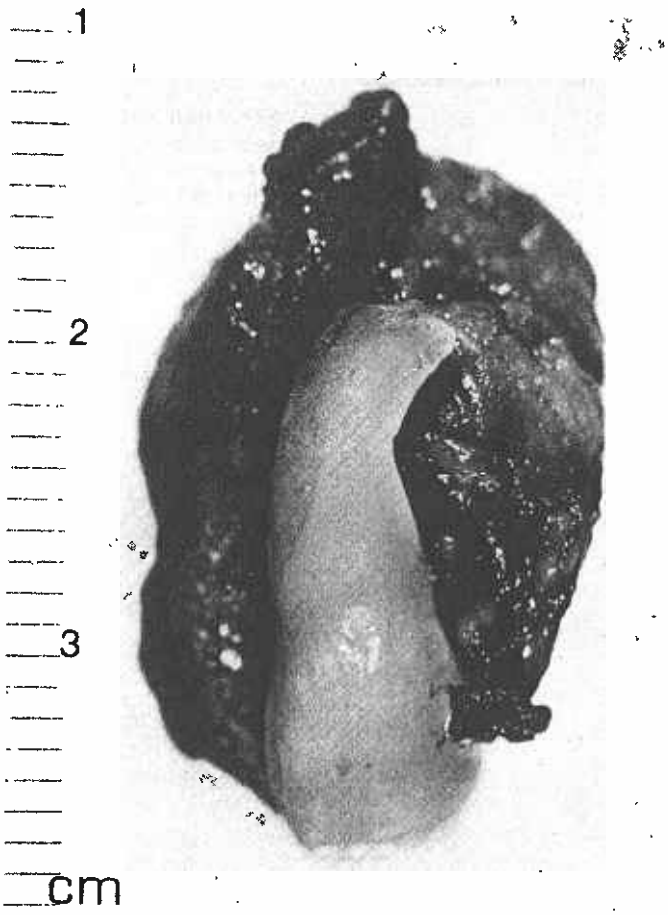


Fig. 2 Gross appearance of an adenomatoid odontogenic tumour associated with an unerupted tooth

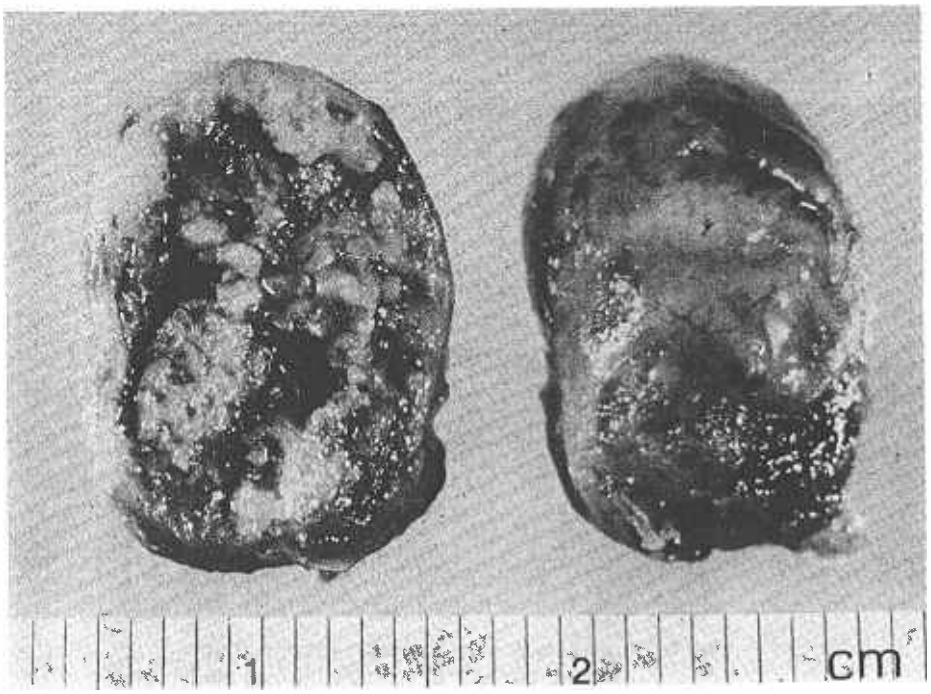


Fig. 3 Gross appearance of the granular content of an adenomatoid odontogenic tumour

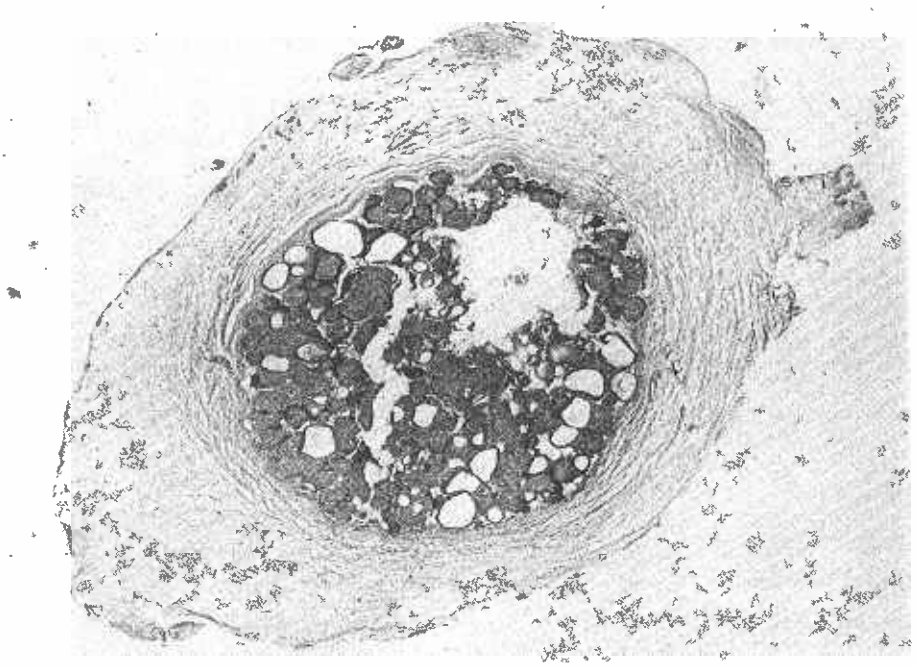

Fig. 4 Low power view to show a solid adenomatoid odontogenic tumour. Note the thickened fibrous capsule (Haematoxylin and eosin stain. Original magnification × 7)

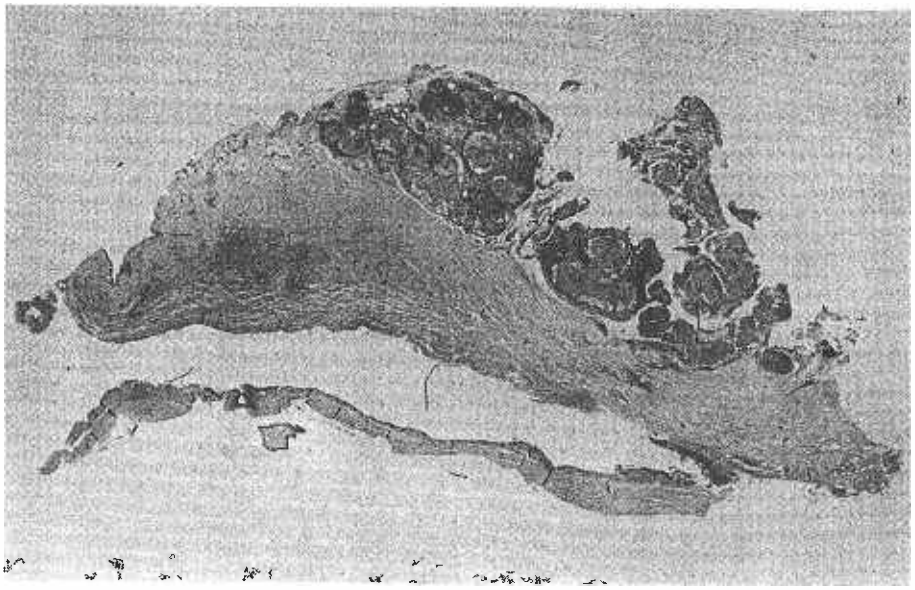

Fig. 5 Low power view showing focal tumour masses backed by a thick fibrous wall. (Haematoxylin and eosin stain. Original magnification × 7)

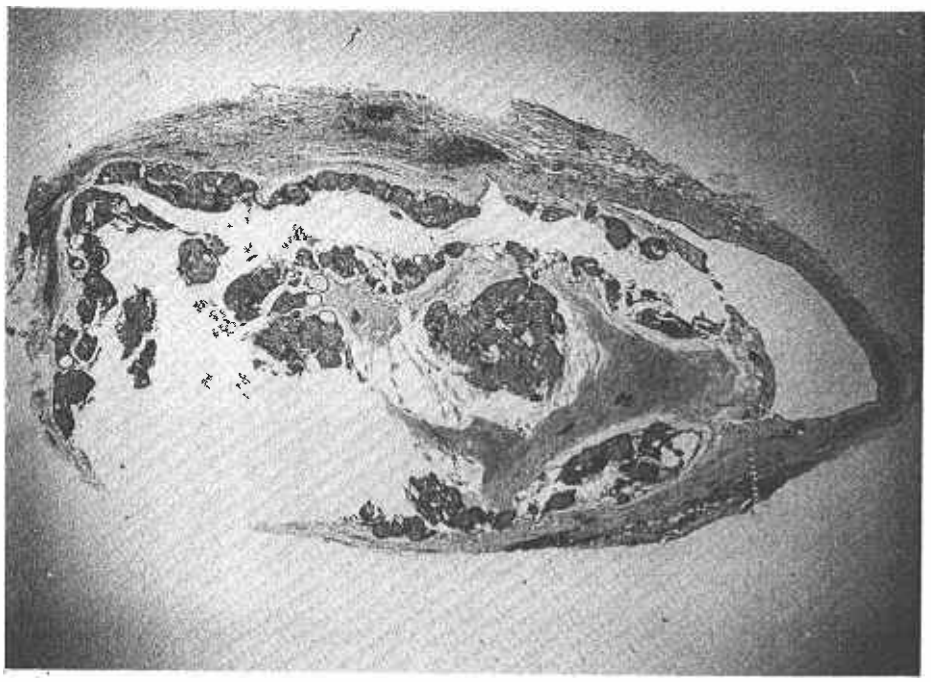

Fig. 6 A cystic adenomatoid odontogenic tumour with the tumour masses forming a mural lining (Haematoxylin and eosin stain. Original magnification × 7)

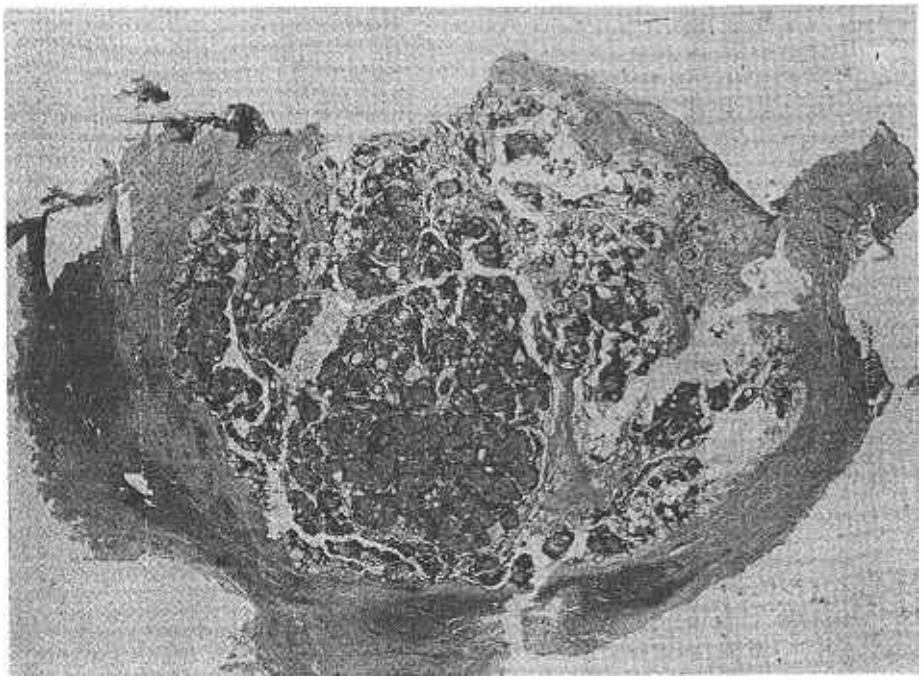

Fig. 7 A solid adenomatoid odontogenic tumour consisting of locules of tumour tissue separated by fibrous septa. (Haematoxylin and eosin stain. Original magnification × 7)

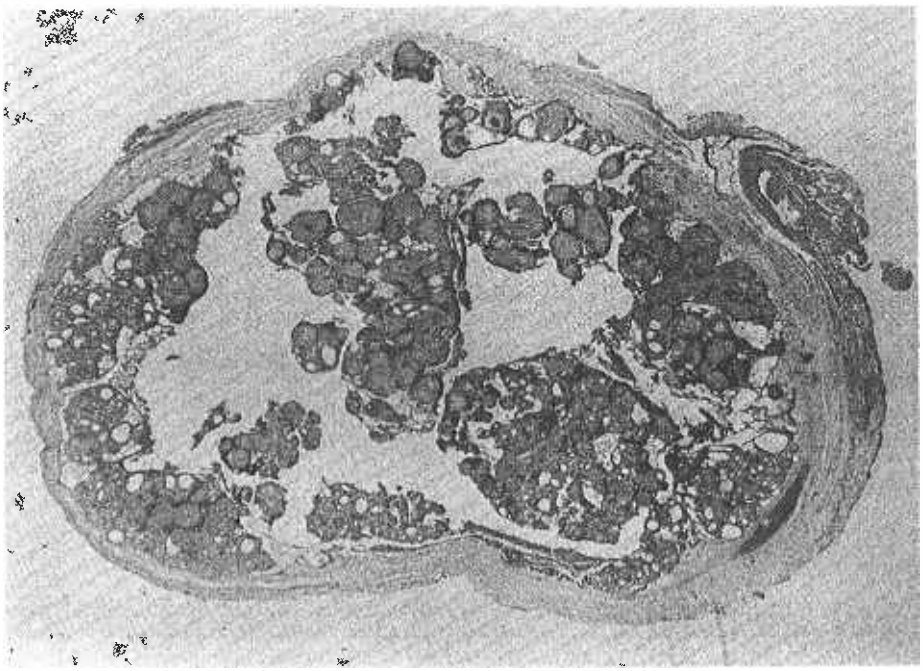

Fig. 8 Marked luminal proliferations obliterating the cystic space (Haematoxylin and eosin stain. Original magnification × 7)

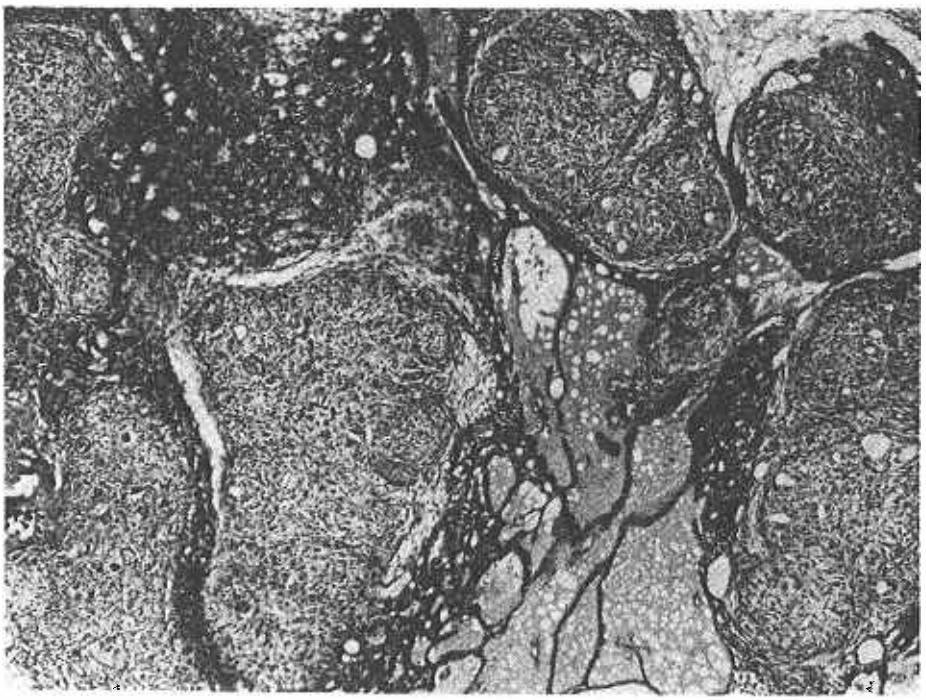

Fig. 9 Ameloblast-like cells forming sheets, nests and cords in a scanty vascular stroma (Haematoxylin and eosin stain. Original magnification × 40)

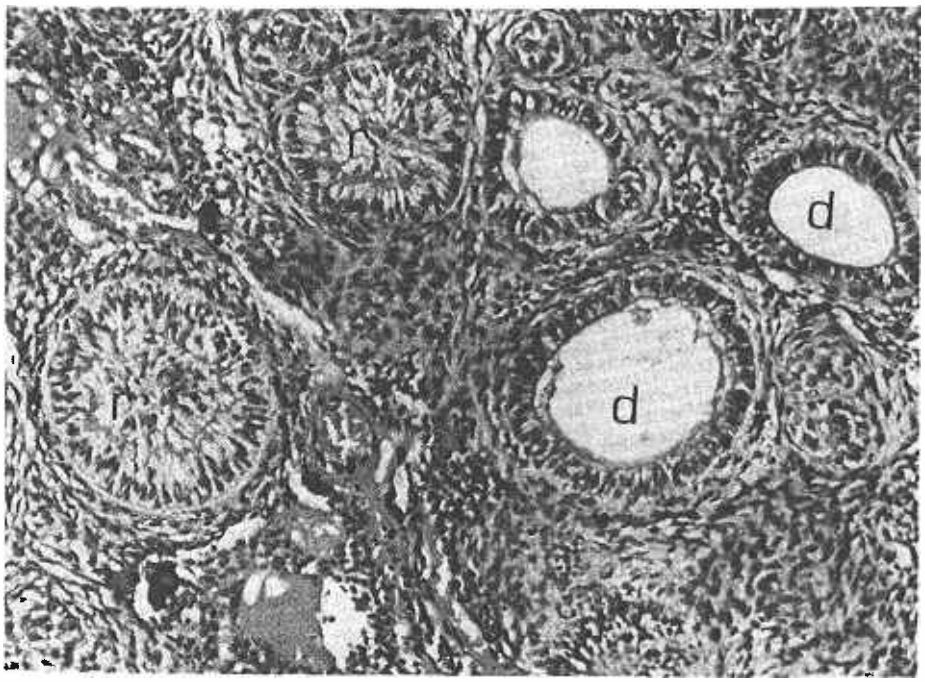


Fig. 10 Tall columnar cells forming an outer row in rosettes (r) or lining duct-like structures (d). Note a single calcific focus at the junction of the tumour epithelium and stroma (left corner). (Haematoxylin and eosin stain. Original magnificiation × 100)

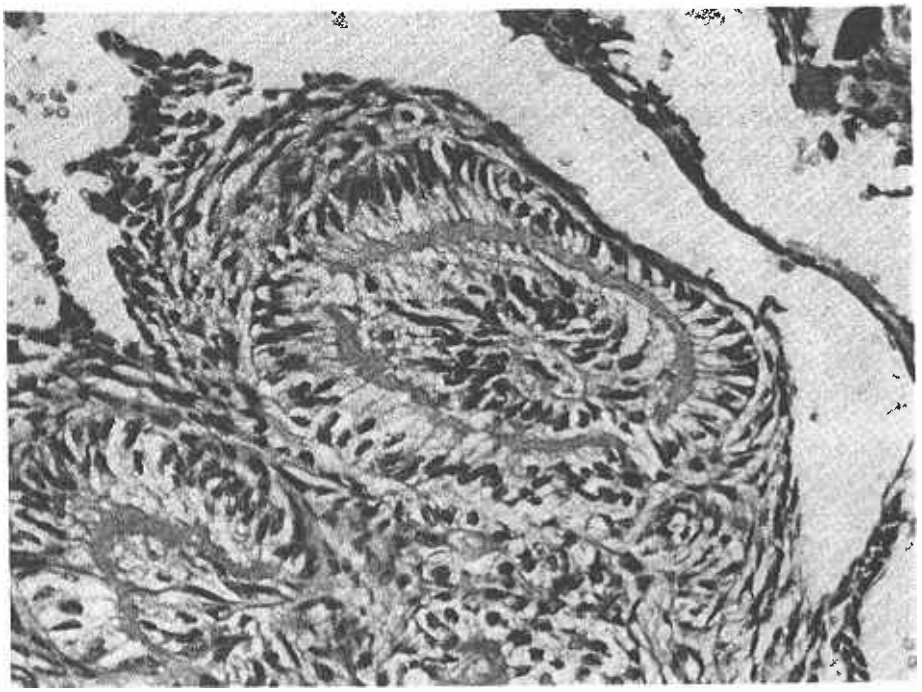

Fig. 11 Details of the eosinophilic deposit between opposing rows of cells in rosettes. (Haematoxylin and eosin stain. Original magnification × 400)

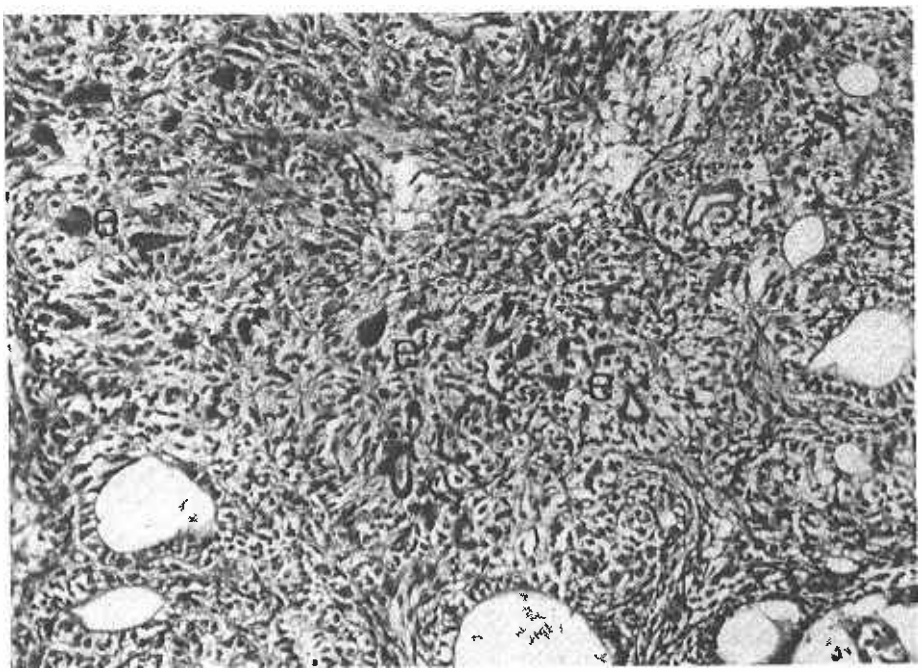

Fig. 12 Eosinophilic droplets (e) may be found scattered throughout the solid tumour areas. (Haematoxylin and eosin stain. Original magnification × 100)

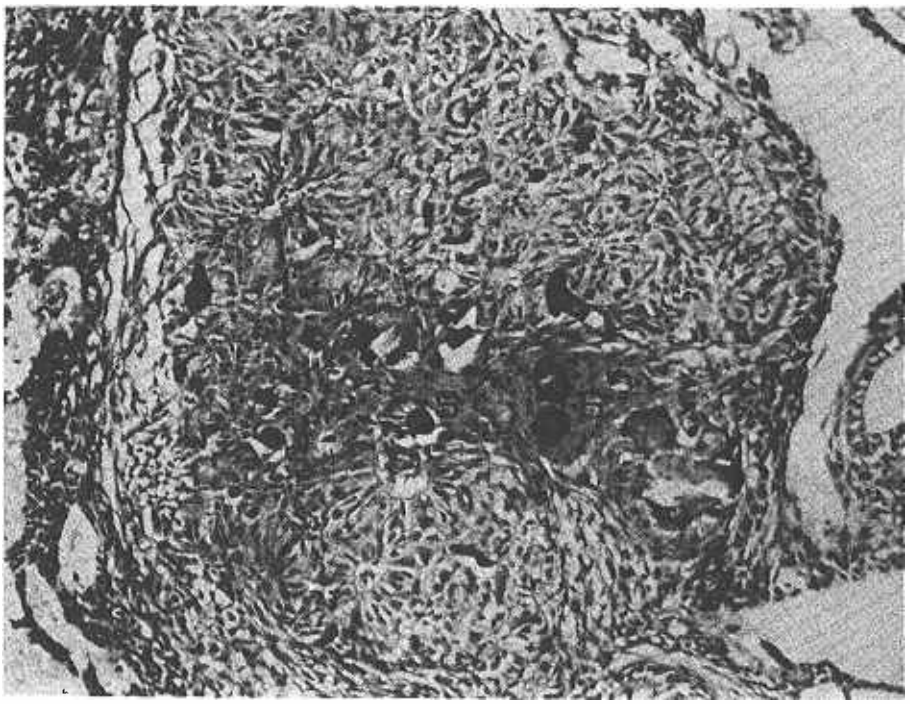

Fig. 13 Spheroidal calcific foci (s) irregularly disposed in the tumour masses. (Haematoxylin and eosin stain. Original magnification × 100)

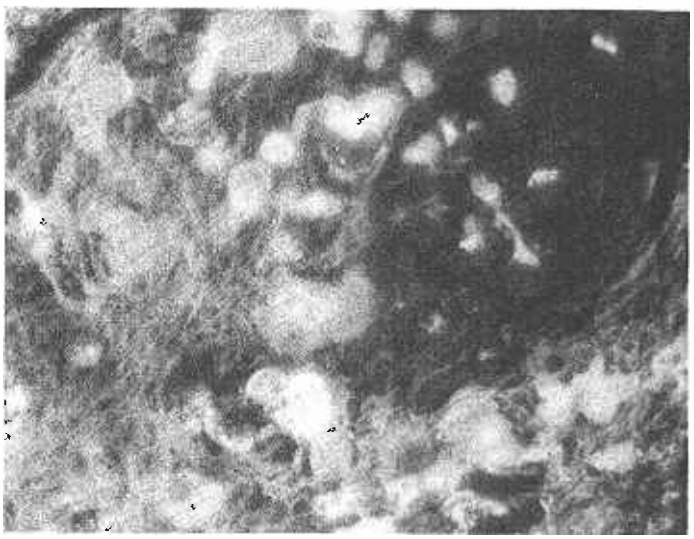

Fig. 14 Fluorescence of calcific foci (Thioflavine T. Original magnification X 100)

## DISCUSSION

Although the histological features of the AOT are well-recognised, this lesion continues to be confused and mis-diagnosed as some other entities notably ameloblastoma and salivary gland tumours. From the historical viewpoint, Ghosh originally refer to this lesion as an "adamantinoma of the upper jaw" (2). Subsequently Bernier and Tiecke (3) proposed the name "adenoameloblastoma" because of its adenoid configuration and resemblance to the ameloblastoma. They and a few other workers viewed it as a variant of the ameloblastoma (3,14,15). Then as more cases were reported in the literature, it became apparent that the clinical and histological features of the AOT as well as its benign, harmartomatous biological behaviour are distinctive and characteristic of this entity, and bears little resemblance to the ameloblastoma which is a more aggressive neoplasm. For this reason, later workers advocated the usage of the term adenomatoid odontogenic tumour in preference to adenoameloblastoma so as to ovoid misinterpretation and errors in treatment (11,15,16). This view was also adhered to in this present study. In the WHO monograph on Histological Typing of Odontogenic Tumours, Jaw Cysts and Allied Lesions, both terms are used (13).

Apart from nomenclature and terminology as a cause for misinterpretation, the histological features of the AOT though classical to the trained eye, could often be misdiagnosed as some other pathology. This is especially true when small incisional biopsy specimens are submitted for histological examination. The relative uncommon occurrence of this lesion combined with the fact that the main tumour cell type in both the AOT and ameloblastoma is the ameloblast-like cell could also account for the misdiagnosis. However with thorough examination of adequately-sized tissue sample, the histological characteristics of the AOT become immediately apparent. Furthermore additional features like duct-like spaces, scattered intercellular eosinophilic droplets and thioflavine T-positive calcific foci may assist in differentiating an AOT from the ameloblastoma.

Although the duct-like structures do not represent an invariable finding in an AOT, their occurrence have in the past prompted diagnoses including cylindroma (11) and unusual pleomorphic adenoma (18). As the AOT are more commonly found in the anterior maxilla, the lesions could be further misinterpreted as tumours originating from the minor salivary glands of the palate. However differentiation of an AOT from a salivary gland tumour can be resolved on clinical and histological grounds. Clinically, the differences in the ages of the patients at onset, sites of occurrence, behaviours, and recurrence potentials for these two neoplasms should enable a distinction to be drawn quite easily. Histologically, as the main tumour cell type in the AOT is the ameloblast-like cell, this tall columnar cell can be differentiated from the uniform, ovoid cells with darkly stained nuclei as in the adenoid cystic carcinoma and the polyhedral or spindle-shaped myoepithelial cells of the pleomorphic adenoma. Furthermore myxochondroid areas and hyaline cells which may be found in the latter tumour are not seen in an AOT. Though a pseudoencapsulation may be seen in a pleomorphic adenoma or adenoid cystic carcinoma the thick well-formed capsule of an AOT is seldom encountered in these two tumours.

## ACKNOWLEDEMENTS

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