

A UNIQUE CASE OF UNICYSTIC AMELOBLASTOMA WITH DISTINCTIVE MUCOUS METAPLASIA

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Abstract:

We report a case of unicystic ameloblastoma with mucous cell differentiation in the anterior maxilla who suffered from painful swelling for 2 months. A radiograph showed a irregular radiolucent lesion between the roots of 21, 22 and 23. Microscopic examination revealed the cystic lesion was lined with ameloblastic epithelium and goblet cells in the epithelium. The mucous cells reacted positively to PAS stain. The possible pathogenic mechanism of this case reflects the pluripotential character of the odontogenic epithelium. The prognosis is probably that expected for conventional unicystic ameloblastoma. Significance of identifying such case of mucous metaplasia in unicystic ameloblastoma is that it should be differentiated from mucous cell containing lesions like glandular odontogenic cyst and mucoepidermoid carcinoma.

Key words: Unicystic ameloblastoma, Prosoplasia, mucous metaplasia

INTRODUCTION:

Ameloblastoma is benign tumor of odontogenic epithelial origin. It accounts for ~1% of all odontogenic cysts and tumors¹. There is no significant difference in incidence between males and females. It most frequently involves the posterior region of lower jaw and has very wide age range (10-92 years)¹. Although it is slow growing it can show aggressive behavior. Radiographically, the lesion always presents with multilocular or unilocular radiolucency, commonly accompanied by the resorption of the tooth roots. It has been studied extensively because of its unique clinicopathological features.

According to recent 2017 WHO classification ameloblastoma can be of four types i.e., conventional, unicystic, extraosseous/peripheral type and metastasizing type. For conventional ameloblastoma two predominant growth patterns are follicular and plexiform. Conventional ameloblastomas also have some less common cell types, including acanthomatous, granular, and basaloid. Additionally, other rare cell differentiations, such as mucous cell differentiation,

have been documented in the literature. Mucous metaplasia is commonly seen in various odontogenic cysts. However its occurrence in ameloblastoma is extremely rare phenomena. Until now only nine such cases have been reported in the literature (table 1). Out of which only two cases were associated with unicystic ameloblastoma. The present paper reports third case of unicystic ameloblastoma with mucous metaplasia.

Significance of identifying such case of mucous metaplasia in unicystic ameloblastoma is that it should be differentiated from mucous cell containing lesions like glandular odontogenic cyst and mucoepidermoid carcinoma which is discussed later in this article.

CASE REPORT :

A 24-year-old male patient reported with pain and swelling on left side of anterior palate region since one and half month. For which he visited hospital and got Root canal opening done with 21 22 and 23 but still swelling didn't subsided.

COMPARISON OF ALL CASES WITH MUCOUS CELL DIFFERENTIATION

S.no.	Author	Age/sex	Location	Diagnosis
1.	Hartenian et al ² 1976	53/F	Anterior mandible	Solid multicystic ameloblastoma
2.	van Wyk et al ³ , 1986	21/F	Anterior maxilla	Unicystic ameloblastoma
3.	Raubenheimer et al ⁴ ,1995	Not available	Anterior mandible	Follicular ameloblastoma
4.	Takata et al ⁵ , 1999	51/M	Anterior maxilla	Desmoplastic ameloblastoma
5.	Wilson et al ⁶ , 2001	31/M	Anterior mandible	Solid multicystic ameloblastoma
6.	Punnya et al ¹ 2008	17/M	Anterior mandible	Solid multicystic ameloblastoma
7.	Punnya et al ¹ 2008	32/M	Anterior maxilla	Desmoplastic ameloblastoma
8.	Yoon et al ⁷ 2009	24/M	Posterior mandible	Unicystic ameloblastoma
9.	Gata et al ⁸ 2015	80/M	Posterior mandible	Solid multicystic ameloblastoma
10.	<u>Present case</u>	<u>24/M</u>	<u>Anterior maxilla</u>	<u>Unicystic ameloblastoma</u>

Intraorally swelling was 3×2 cm in size extending from gingiva of 21, 22, 23 and 24 to mid-palatine raphe with roughly oval shape (fig.1 A&B) Margins were ill-defined. Radiograph shows ill-

defined radiolucency involving root of 21, 22 and 23(fig.2 A&B). Enucleation of cystic lesion with apicectomy of 21, 22 & 23 was done.

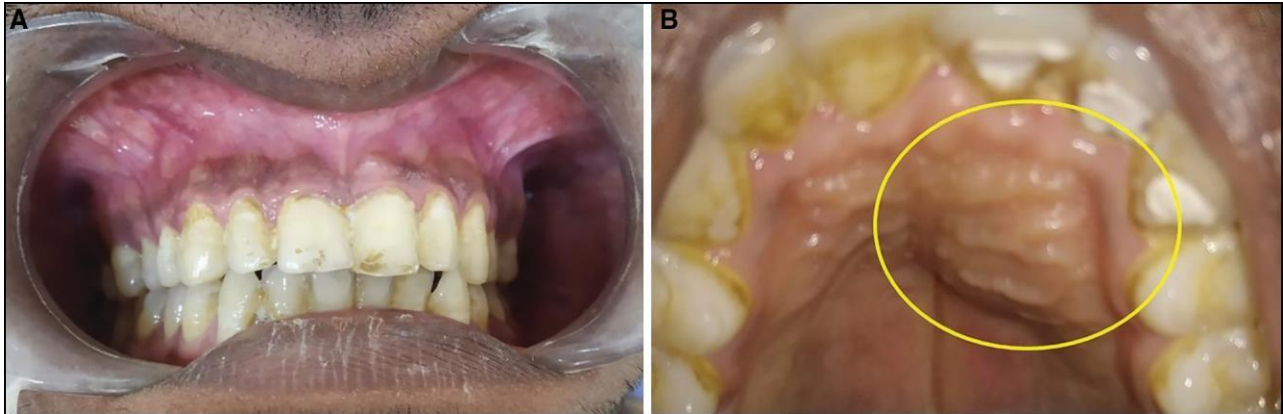


Figure 1: (A) No swelling in buccal vestibular region. (B) Diffused swelling in palatal gingiva of 21, 22, 23, and 24 upto midpalatine raphe

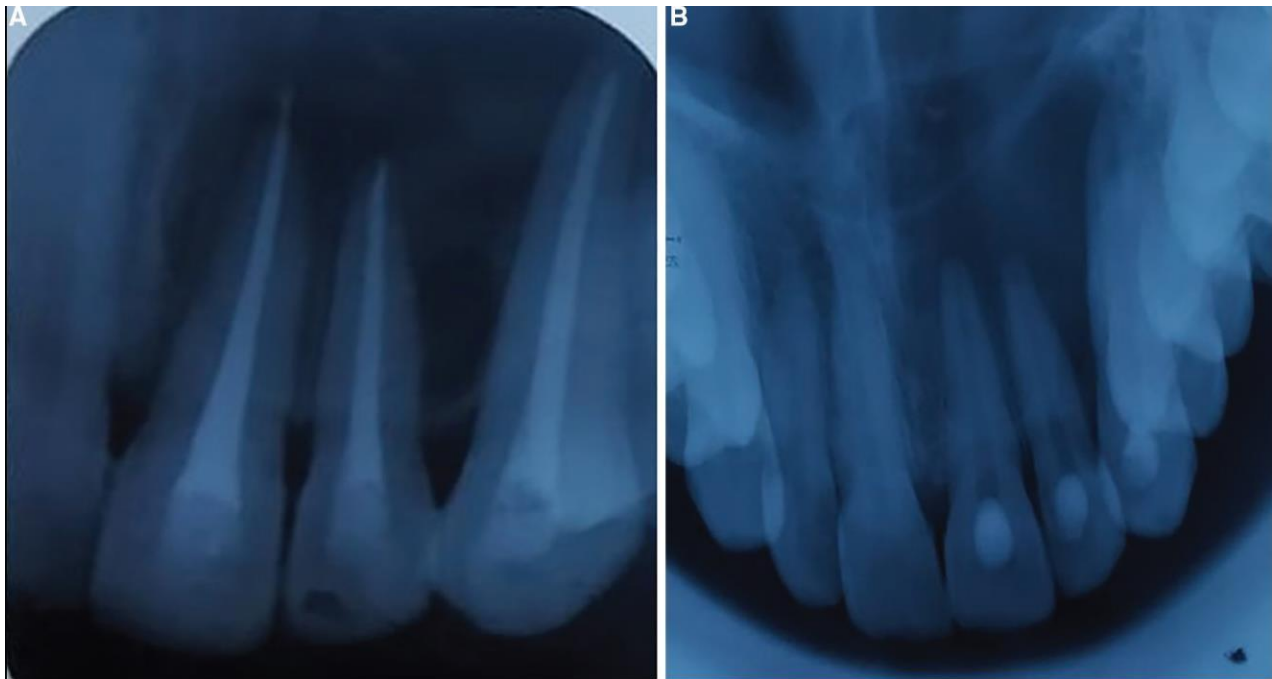


Figure 2: (A and B) Ill-defined radiolucency extending from mesial aspect of root of 21 to mesial of root of 23

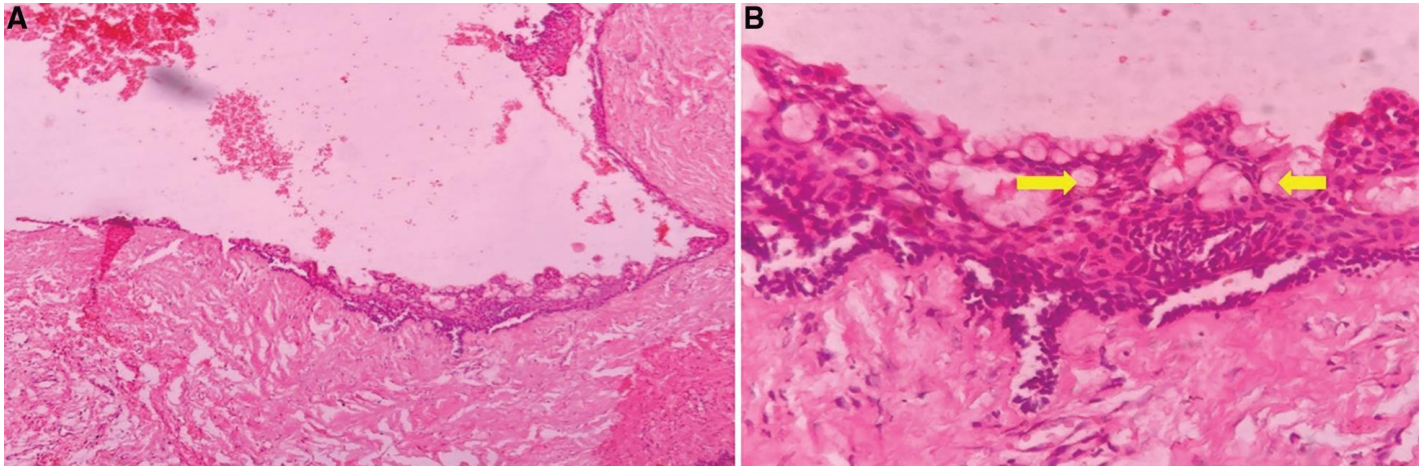


Figure 3: (A) Cystic cavity focally lined by the nonkeratinized stratified squamous epithelium. (B) Superficial cell layers of lining epithelium 37 also showed mucous metaplasia

H & E stained section of cystic lesion showed cystic cavity focally lined by non-keratinized stratified squamous epithelium of variable thickness. At places lining epithelium exhibited basal cuboidal to columnar cells with hyperchromatic nuclei and loosely arranged stellate reticulum like cells suggestive of odontogenic epithelium. Focally superficial cell layers of lining epithelium also exhibited mucous metaplasia. Fibrous connective tissue wall exhibited epithelial islands in form of follicles. These follicles showed peripheral tall columnar cells with hyperchromatic nuclei and central stellate reticulum like cells. Connective tissue capsule shows parallelly arranged collagen bundles peripherily with mild inflammatory infiltrate, moderate degree vascularity and areas of hemorrhage (fig 3 A & B)

Therefore final diagnosis was given as Unicystic Ameloblastoma – Mural variant with mucous metaplasia.

DISCUSSION:

As stated by Robinson, Ameloblastoma is usually unicentric nonfunctional, intermittent in growth, anatomically benign and clinically persistent. Historically, Ameloblastoma has been recognized for over a century and a half. Its frequency, persistent local growth, and ability to produce marked deformity before leading to serious debilitation probably account for its early recognition. Recurrence, especially after conservative treatment, has also contributed to the awareness of this lesion²

Histologically conventional ameloblastoma shows various types like follicular, plexiform, acanthomatous, Granular, desmoplastic and basal cell. In granular cell ameloblastoma granularity is due to marked transformation of the cytoplasm of stellate reticulum like cells into a coarse, granular eosinophilic appearance previously it was considered to be aggressive in nature, with a marked propensity for recurrence and metastasis. But Recently it is considered that all types of histological variants of conventional ameloblastoma exhibits similar aggressive behavior. So treatment modality is similar in all variants².

Ameloblastomas have been broadly divided into three biological variants, of which the unicystic type is least aggressive⁹. Unicystic ameloblastoma shows an incidence of around 5–22%, but with a lower recurrence (6.7–35.7%) generally affecting a younger population³. It presents three histopathologic subtypes. The luminal variant is a cystic lesion with a flat ameloblastic cystic lining. The intraluminal subtype is characterized by tumor growth into the cyst lumen, while the mural subtype presents infiltrating growth into the wall of the cyst and possibly beyond into the surrounding bone³.

Leider et al¹⁰ proposed three pathogenic mechanisms for the evolution of UA: the reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development; ameloblastomas arise in dentigerous or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining; and a solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts and develops into a unicystic lesion¹¹.

Ameloblastomatous epithelium of UA can show various histologic patterns most commonly plexiform pattern. Some cases are reported with granular, acanthomatous and basaloid pattern in lining epithelium³. Mucous cell differentiation is also seen in some cases. Present article reports a tumor which exhibits characteristic histopathological appearance of Unicystic ameloblastoma. Along with this most interesting aspect of this tumor is presence of mucous cell differentiation⁵.

The finding of clear or pale cells such as mucous cells—in ameloblastomas is very rare, but not unreported, phenomenon. But its presence in

odontogenic cysts like radicular cyst is common¹². The diagnostic implications of the presence of clear or pale cells in ameloblastomas are several and relate mainly to the question of histopathologic differential diagnosis^{13,14}. Because of presence of mucous cells there are possibilities of other histopathological diagnoses like mucoepidermoid carcinoma, clear cell odontogenic tumor or clear cell odontogenic carcinoma.

The pathogenesis of mucous cells within ameloblastomas is not clear. It has been suggested that the odontogenic epithelial component might differentiate ability⁷ and mucous cell differentiation of the epithelial cells might be a response to an altered environment, such as inflammation or necrosis in salivary gland tumors¹⁵. Another hypothesis is that the differentiated cells undergo the process of prosoplasia, which is known as a forward differentiation either to a higher or intricate function or to a more complex level of organization. The most classical example is the differentiation of the squamous epithelial cells to mucous cell¹⁶. However, those hypotheses need to be further explored in more cases of ameloblastoma with mucous cell differentiation in the future.

Whether a unicystic ameloblastoma in association with mucous cells represents a collision growth of two distinct components or a metaplastic phenomenon within a unicystic ameloblastoma remains speculative. It was previously suggested that the epithelial lining of the odontogenic cysts can undergo metaplasia from a stratified squamous

to a more highly differentiated ciliated columnar or glandular type. The stimulus for such metaplasia is unclear, although increased hydrostatic pressure has been suggested¹⁷. The possible pathogenic mechanism of this case would appear to be a reflection of the pluripotential character of the odontogenic epithelium. This case indicates that

multipotential odontogenic epithelial tissue has the ability to develop diverse differentiation. The prognosis in this case will probably be as expected for conventional unicystic ameloblastoma.

The histopathologic differential diagnosis due to presence of mucous cells includes salivary gland tumor, especially a mucoepidermoid carcinoma, which is known to occur commonly in the jaws, and glandular odontogenic cyst. The diagnosis of mucoepidermoid carcinoma is based on identification of mixture of mucous, intermediate, and epidermoid cells, while glandular odontogenic cyst (GOC) is predominantly cystic lined by a typically thin stratified squamous epithelium with characteristic superficial layer of cuboidal/columnar cells which occasionally exhibit cilia^{6,18}. Additionally, the epithelium in GOC may show swirling spherical aggregates, papillary proliferation, and pools of mucin^{1,18}.

CONCLUSION:

The existence of mucous cells in ameloblastoma thus illustrates the multipotentiality of odontogenic epithelium and highlights the need for careful histopathologic assessment of ameloblastoma for documentation of such extraordinary heterogeneities in its histopathologic spectrum and prognostic significance, if any, of this rarity.

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