UNICYSTIC AMELOBLASTOMA WITH DENTINOGENESIS: A RARE CASE REPORT

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Abstract:

Ameloblastoma is one of the most common odontogenic tumors with benign but aggressive clinical features. It represents 1% of all tumors of jaw bone. Though it is characterized by wide variety of histologic appearance, it does not generally show evidence of induction in the connective tissue.

Here we report of case of unicystic ameloblastoma in 28 year old female, which showed

evidence of induction in the form of dentinoid and tubular dentin formation.

Keywords: Ameloblastoma, Dentinoid, Induction.

Introduction:

Of all the swellings in the oral cavity, 9% are odontogenic tumors and within this group ameloblastoma accounts for 1% of lesions and has received considerable attention due to its frequency, clinical subtypes and high tendency to infiltrate and recur. Based on the type of odontogenic tissue involved in the process of development, WHO classified ameloblastoma into the group of benign odontogenic epithelial tumors without odontogenic ectomesenchyme. Although it presents microscopically with wide variety of appearance, it does not generally show evidence of induction and no hard tissue formation except in odontoameloblastoma which is classified under separate group [1].

However rare cases of ameloblastoma are reported showing evidence of dentinoid induction by tumor cells but without evidence of active odontogenic ectomesenchyme or presence of dental papilla like tissue. Such cases create doubt regarding histogenesis and cause diagnostic nosologic problem as conventional classification of odontogenic tumors do not apply to these tumors.

Here, we report a case showing features of unicystic plexiform ameloblastoma along with dentinoid and tubular dentin formation and discuss the histogenesis.

Case report

A 28 year old female patient reported to the outpatient department of Oral Medicine &

Radiology, with the chief complaint of asymptomatic swelling in the left mandibular posterior region .According to the history of presenting illness patient had a similar swelling one year back for which she underwent surgical procedure along with the extraction of posterior teeth of the same quadrant. However patient was not having any records of the previous treatment

Extra-oral examination revealed slight facial asymmetry due to presence of diffuse swelling over posterior left mandibular region which was firm and non-tender on palpation. (Fig 1)

Intraoral examination revealed presence of a diffuse swelling extending antero-posteriorly from 32 to retro-molar region. Color was similar to surrounding normal mucosa. Swelling was soft to firm in consistency and non-tender on palpation. (Fig. 2)

A small horizontal erythematous soft tissue growth measuring approximately 2x0.3cm was seen lingual to 32 and 33 extending up to 42. 31 & 41 were missing. (Fig. 3).

The panoramic radiograph revealed a well-defined multilocular radiolucency within the left mandibular body with scalloped borders and thinned out cortical margins extending antero-posteriorly from 32 till ascending ramus. Root resorption of 34 & 35 was well appreciated. (Fig 4) Mandibular occlusal radiograph revealed presence of multilocular radiolucent lesion involving 34 and 35 and extending posteriorly. Expansion and perforation of lingual cortical plate was well appreciated. (Fig.5)

Haemtological and urine investigations were within normal limits. Based on clinical and radiographic findings, a provisional diagnosis of ameloblastoma, recurrence of

previously treated odontogenic cyst/tumour, was made and incisional biopsy was performed.

The microscopic examination of H & E stained section revealed presence of cystic lumen lined by epithelium and surrounded cells with polarized, palisaded, hyperchromatic nuclei and vacuolization basally, fulfilling the Vicker & Gorlin *(V&G)* criteria. At focal areas the cells are cuboidal. (Fig 6) At few areas, intraluminal proliferation of lining epithelium in plexiform pattern is seen. (Fig 7) A part of section showed mural growth in the form of follicle of ameloblastoma growing in connective tissue capsule.

In between the anastomosing cords of plexiform ameloblastomatous proliferation, dentinoid formation along with sheets of tubular dentin was well appreciated.(Fig 8) Sheets of tubular dentin by thin capsule. The cystic lining showed presence of odontogenic epithelium consisting of basal cell layer resembling ameloblasts and 2 -3 layers of stellate reticulum like suprabasal cells. Basal cell layer revealed presence of columnar were also seen in the cystic lumen.(Fig 9)

The capsular connective tissue stroma was fibrocellular with moderate inflammatory cell infiltrate chiefly lymphocytes.

In the deeper connective tissue showed lots of giant cells along with cholesterol clefts. (Fig 10) Dentinoid material stained positively with the Van Gieson stain. (Fig 11)

Discussion:

Odontogenic tumours originate from the tissues of tooth forming apparatus and reproduce to a greater or lesser extent inductive interrelationship between the various parts of the normal tooth germ. They

constitute a very diverse group of lesions due to the different degrees of inter-tissue interaction and various growth patterns[2].

Very few cases are reported in the literature. In 1972 Dunlap and Fritzlen reported the presence of tubular dentin in duct like or glandular structures of an adenoameloblastoma [3]. The next mention of a similar lesion was by Orlowaski *et al* cribriform proliferation together with

Then, Brannon first coined the term adenoid ameloblastoma with dentinoid while reporting a case of similar neoplasm with recurrence potential from Armed Forces Institute of pathology-USA. Brannon RB (1994) further stated that Adenoid ameloblastoma with dentinoid can be considered a variant of ameloblastoma. It has histologic features of both ameloblastoma and AOT, including calcified tissue, but should be considered a

presence of ghost cells and dentinoid material. [5] Slabbert *et al* (1992) also reported a case of ameloblastoma with dentinoid formation. [1]

(1991) and they interpreted the lesion as plexiform ameloblastoma with dentinogenesis. [4]

Tajima et al (1992) reported a case showing ameloblastomatous adenoid neoplasm with decided potential for extension and recurrence as cited by Evans[6].

In our case the clinicoradiographic presentation and histopathology suggested the diagnosis of unicystic plexiform ameloblastoma with dentinogenesis.

Unicystic ameloblastoma present on an average a decade earlier than the solid multicystic Ameloblastoma which justifies the occurrence in our case in 3rd decade [7]. Radiographically our lesion presented with a multilocular pattern which is though rare, is

not uncommon for unicystic Ameloblastoma. Eversole et al identified multilocular appearance as one of the six radiographic patterns for UA's [8].

Histologically, there were plexiform ameloblastomatous luminal growth patterns interspersed with eosinophilic dentinoid globules and sheets of tubular dentin

Slabbert *et al* did transmission electron microscopic study and stated that the dentinoid was seen to consist of interlacing fibrils 40 nm in diameter, consistent with collagen showing faint periodicity of the fibres. The fibres[12, 13, 14] were separated by a structureless ground substance and showed parallel layering at the edges. [1]

Presence of cuboidal cells consistent with odontoblasts, lining the dentin like material on one side is also reported by Allen *et al* [9] and Orlowski *et al*. [4]

So it can be hypothesized that in atleast some of these lesions, the odontogenic epithelial cells induce the stromal cells to develop into odontoblasts

According to Pindborg *et al* dentinoid is the result of odontogenic

In our case one can easily appreciate the presence of bipolar and stellate shaped cells entrapped in the dentinoid globules which seem to be entrapped odontoblasts. (Fig 8)

Presence of such entrapped cells was also reported by Dunlop and Fritzlen and Slabbert *et al* which show presence of some cells in the homogenous dentinoid material. and he reported that dentinoid was deposited by mesenchymal cells .[1, 3]

and these induced cells lay down the dentinoid like material.

epithelial induction on more mature connective tissue [10, 11]. Initiation induction for dentinogenesis requires epithelial - mesenchymal interaction, the mechanism of which is not clearly understood.

Van Wyk *et al* have done microscopic and ultrastructural study of the epithelial - connective tissue interface of the ameloblastic fibroma with dentinoid formation and concluded that the features seen at the epithelial connective tissue interface represent a spectrum of abortive inductive features. Van Wyk's observations also revealed entrapped cells in the dentinoid material and they proposed that some of these entrapped cells are aborted odontoblast. They further stated that, epithelial differentiation – from odontogenic epithelium to cylindrical ameloblast like cells is needed to progress to a stage from amorphous hyaline like material to dentinoid epithelio-mesenchymal cells may even reach advanced stages of maturity with matrix deposition. They (Papagerakis *et al)* concluded that the protein production of the odontogenic epithelium is not restricted to that of enamel proteins.

Papagerakis *et al* demonstrated that ameloblastic epithelial cells in mixed odontogenic tumors expressed gene products normally present in ectomesenchymal cells and resulted in conversion and co expression of mesenchymal phenotype. They showed that epithelial cells committed to ameloblastic differentiation can express markers of the osteoblast / cementoblast lineage in pathological conditions and the

In our case, some of the stromal microcystic spaces were also seen to be filled with

blood. According to Lucas, in the process of formation of stromal cyst, in the plexiform ameloblastoma, the blood vessels often persist and dilate instead of disappearing,

thus it is likely to represent a purely secondary change and not a neoplastic process.

On the other hand Van Rensberg *et al* advanced the theory that excessive stimulation of angiogenesis during tumor development and trauma such as tooth extraction could be the reason for the development of vascular components. [15,19,20]

In our case development of haemangiomatous microcystic spaces and presence of extravasated red blood cells could have been due to previous surgical intervention.

The presence of cholesterol clefts associated with giant cells was seen at many places in the capsular connective tissue stroma. Cholesterol clefts in radicular cyst have been postulated to be associated with disintegrating red blood cells in a form that readily crystallizes in the tissue. [16, 21, 22]. However the presence of cholesterol clefts with associated giant cells and foamy macrophages (foam cells) is not as common in ameloblastoma/odontogenic lesions as it is in our case. The presence of extravasated blood could have been resulted in the development of cholesterol clefts and giant cells.

After studying all these reported cases, it is observed that, though they present mostly as unicystic ameloblastoma, their behavior is similar to conventional AB with multiple recurrences and wider

excision should be planned for treatment. In our case also since it was a recurrence, treatment of choice for conventional AB was taken into consideration and hemimandibulectomy was done.

This case once again proves that, induction in ameloblastoma, is possible, which suggest that classification of odontogenic tumors on the basis of induction is not justified.

Hence, we conclude the present lesion to be a rare case of plexiform unicystic ameloblastoma with dentinoid which may be under diagnosed. Therefore, all the pathologists should have a thorough understanding of this entity and review the microscopic slides thoroughly and completely in order to make an accurate

diagnosis. Further case reports on plexiform unicystic ameloblastoma with dentinoid are expected to shed a light on the biological behavior and nature of this unique tumor.

In the third edition of WHO histologic typing of odontogenic tumors in 2005, this term is not included probably because of the rarity of the reports. Now, as sufficient number of cases of ameloblastoma with dentinogenesis are reported in the literature, we suggest that it should be included in the next edition of WHO histologic typing of odontogenic tumors

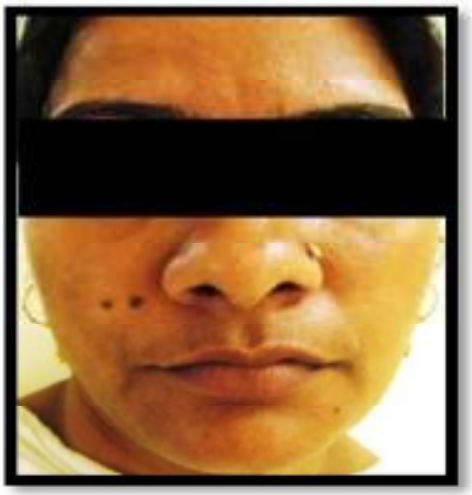

Figure 1: Extraoral photograph showing no discernable extra-oral swelling.

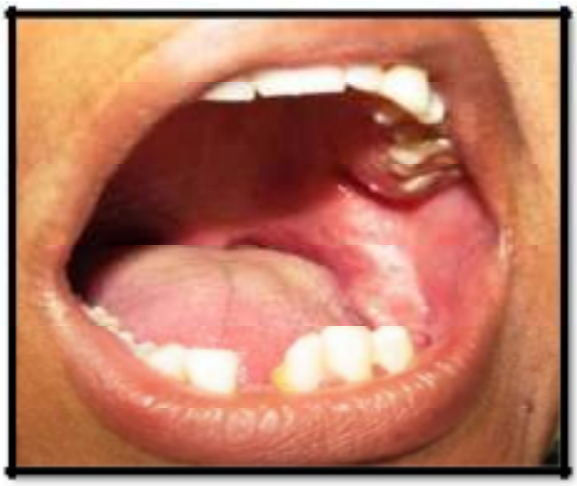

Figure 2: Intraoral photograph showing presence of diffuse swelling posterior to 34

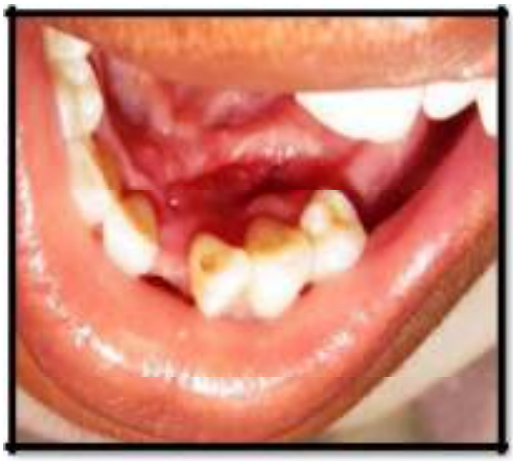

Figure 3: Intraoral photograph showing non-ulcerated erythematous soft tissue growth on the floor of mouth in relation to 32, 33

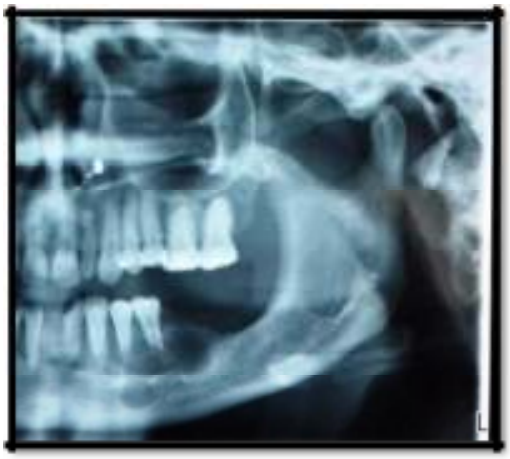

Figure 4: OPG showing well defined multilocular radioluscency apical to 33, 34, 35 as well as in the ascending ramus.

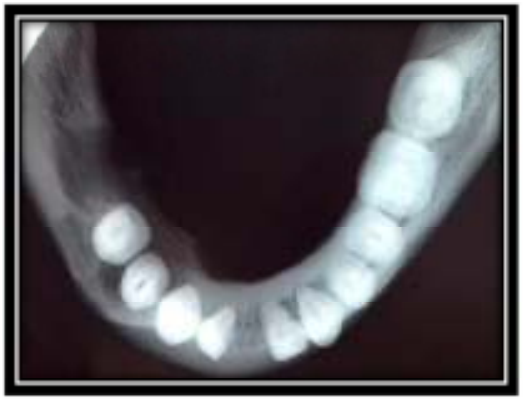

Figure 5: Occlusal radiograph showing multilocular radioluscency involving 34, 35 extending posteriorly along with perforation of lingual cortical plate.

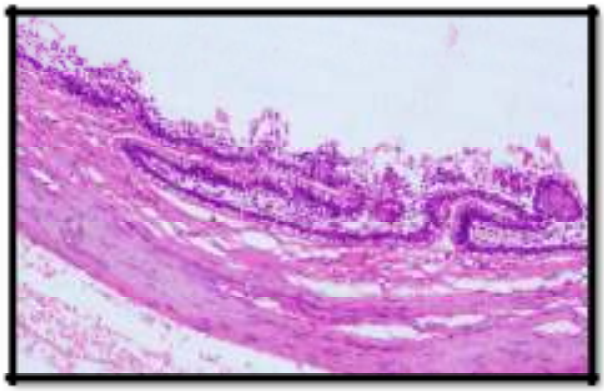

Figure 6: Photomicrograph showing cystic lesion showing proliferated ameloblastic epithelial lining. (*H& E* 100x)

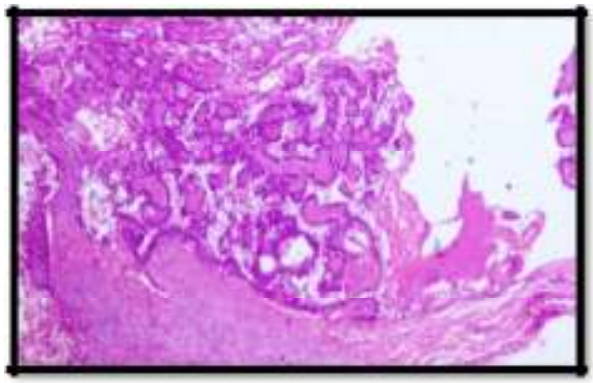

Figure 7: Photomicrograph showing proliferating odontogenic epithelium within the cystic lumen (*H&E* 40x)

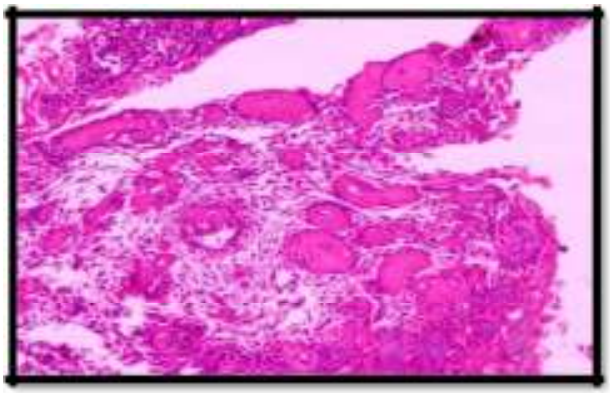

Figure 8: Photomicrograph showing dentinoid formation within the ameloblastomatous proliferation. (*H& E* 100x)

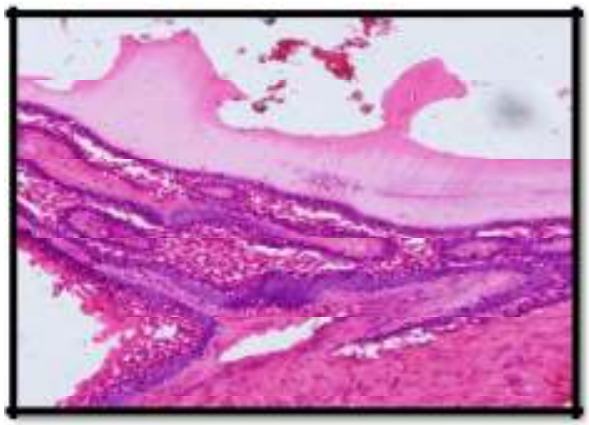

Figure 9: Photomicrograph showing Sheets of tubular dentin deposition. (*H& E* 100x)

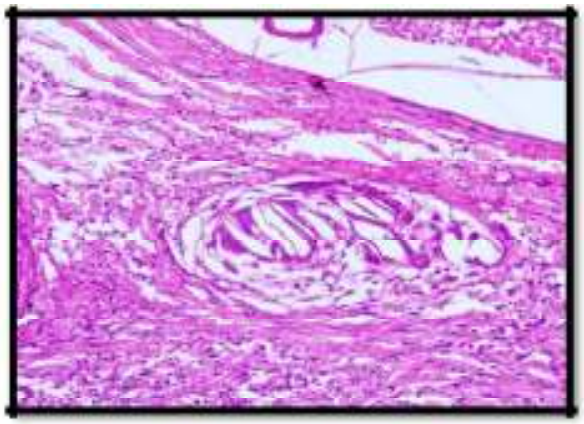

Figure 10: Photomicrograph showing connective tissue capsule showing abundant inflammatory cell infiltrate and cholesterol clefts with giant cells. (*H& E* 100x)

Figure 11: Photomicrograph showing positivity for dentinoid. (*Van Gieson Stain* 100x)

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