原文題目(出處)：	Unicystic Mural Ameloblastoma: An Unusual Case Report. Case Rep Dent 2013, Article ID 957418
原文作者姓名：	V Nagalaxmi, Mithare Sangmesh, Kotya Naik Maloth, Srikanth Kodangal, Vani Chappidi, and Stuti Goyal
通訊作者學校：	Sri Sai College of Dental Surgery
報告者姓名(組別)：	劉奇錚 (Intern I 組)
報告日期：	102.5.6

內文：

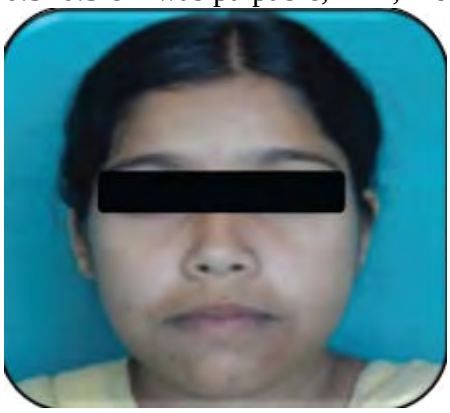
1. Introduction:
   Many benign lesions cause mandibular swellings, and these can be divided into odontogenic and nonodontogenic origin. The most common tumor of odontogenic origin is Ameloblastoma. More than 80% of all Ameloblastoma are solid or multicystic variants. The 20% was shared by unicystic Ameloblastoma and peripheral Ameloblastoma.
2. Case Report:

18-year-old patient

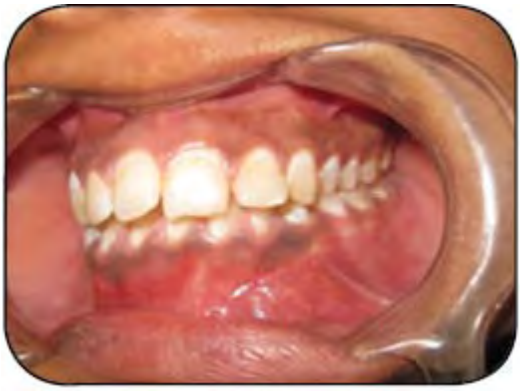
C.C.: swelling in the lower left front teeth region, for 3 months.

P.I.: Patient was apparently well 3 months back and noticed a swelling and displacement of teeth in the left lower front tooth region and reports of having pain in the same region, for 3 months. Pain was of dull aching
type, which was intermitted, and it aggravates on putting mastication and relieves on rest. Pain was not associated with fever and no medication was taken.

Extraoral examination: Size about 4x3 cm, overlying skin was normal, no visible pulsation or discharge was seen, firm, tenderness (+), no local rise of temperature, noncompressible. A single left submandibular lymph node of size measuring about 0.5x0.5 cm was palpable, firm, mobile, nontender.



Intraoral examination: A single diffuse swelling was seen in the mandibular left buccal vestibule. at 31~34 area, size approximately 4x2 cm. No discharge was seen. Lingual cortical plate expansion was seen irt to 31, 32, and 33 (33 missing), firm, smooth surface, nonfluctuant, no pulsations felt. On needle aspiration, brown yellow fluid was aspirated. Diagnosis was given as dentigerous cyst.



3. Differential Diagnosis

Ameloblastoma

Old age, site, multilocularity, impacted lower canine.

Calcifying epithelial odontogenic tumor (CEOT)

Unilocular R/L without R/O flecks.

Odontogenic keratocyst (OKC)

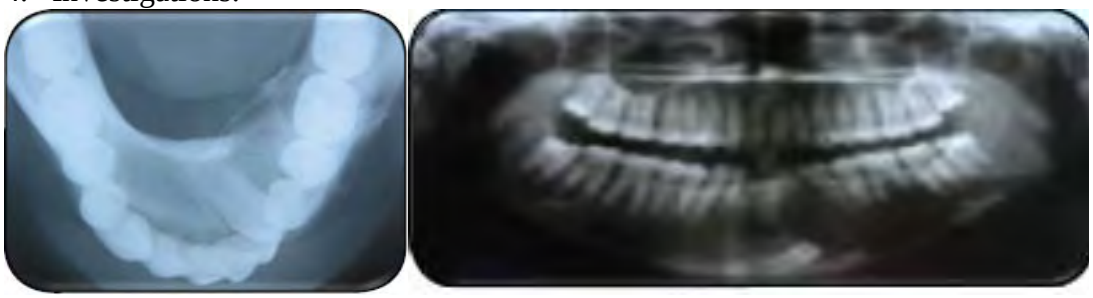
Buccolingual bone expansion ruled out OKC

Central giant cell granuloma (CGCG)

Odontogenic myxoma

Ruled out based on clinical and radiological features

4. Investigations:

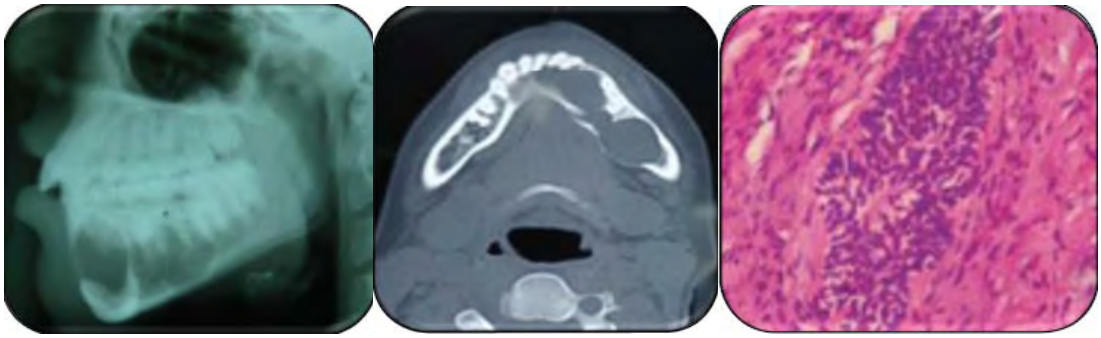


Mandibular occlusal view reveals impacted 33 and expansion of lingual cortical plates irt 32, 33, 34, 35, and 36 and mild expansion of buccal cortical plate.

Panorex shows a well-defined multilocular radiolucency with internal bony septae formation and impacted 33. Displacements of teeth irt 35, 34, 32, 31, 41, 42, and 43 were seen, with root resorption of 36

lateral cephalogram reveals a clear multilocular radiolucency with septae formation

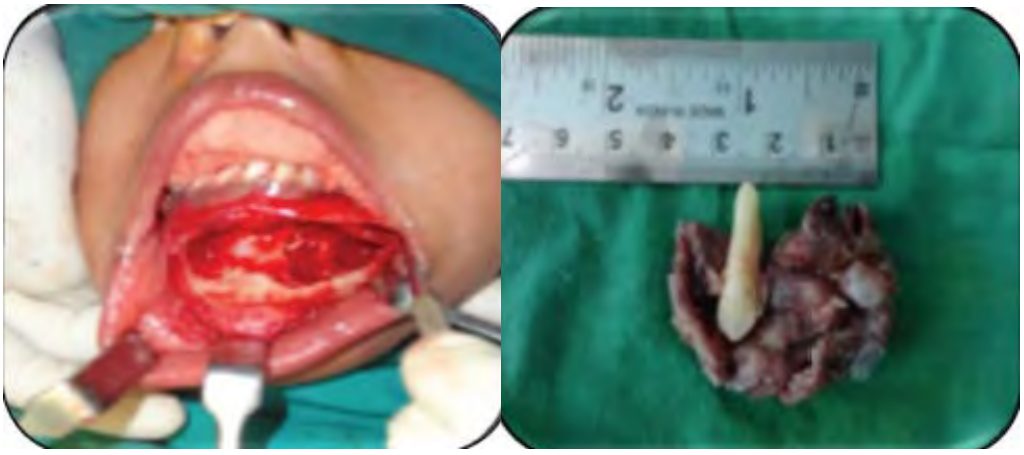
CT scan reveals expansion and perforation of the cortical plates and the extent of lesion



The biopsy tissue shows multiple cystic lesions and collagenous wall of varying thickness. The cyst is lined by odontogenic squamous epithelium of varying thickness with nuclear palisading along the margins and loose stellate reticulum.

They contain moderate to abundant pale acidophilic vacuolated cytoplasm and round to oval vesicular nucleus. There are thin plates of lamellar bone with focal osteoid deposition and patchy areas of hemorrhage. There are focal epithelial invaginations associated with desmoplastic fibrosis.
Based upon the radiological and histopathological report, the case was diagnosed as mural Ameloblastoma.

5. Treatment: Enucleation.



6. Discussion:

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm with variable clinical expression and accounts for 1% of all cysts/tumors of jaws and 18% of all odontogenic neoplasms. It is typically slow growing, locally aggressive and rarely metastasizes but has a high rate of recurrence (55–90%) if not removed adequately.

WHO system of 2003, Ameloblastoma is classified based on differences in biologic behavior, treatment plan and recurrence rate.

(1) Classic solid/multicystic ameloblastoma
(2) Unicystic ameloblastoma (UCA)
(3) Peripheral ameloblastoma
(4) Desmoplastic ameloblastoma, including the so-called hybrid lesions

Unicystic ameloblastoma (UCA) is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas. It usually occurs in a younger age group of 16–20 years, with about 50% of the cases occurring in the second decade of life as in our case. The gender distribution shows a slight male predilection with a male to female ratio of 1.6 : 1. However, when the tumor is not associated with an unerupted tooth, the gender ratio is reversed to a male to female ratio of 1 : 1.8. More than 90% are located in the mandible in the posterior region, followed by the parasymphysis region, the anterior maxilla, and the posterior maxilla. UCA is usually asymptomatic, although a large tumor may cause painless swelling of the jaws with facial asymmetry. Mucosal ulceration is rare but may be caused by continued growth of the tumor. The clinical and radiographic findings in most cases of unicystic ameloblastoma suggest that the lesion is an odontogenic cyst, particularly dentigerous cyst.

Histologic criteria for the diagnosis of unicystic ameloblastoma includes a cyst lined by ameloblastic epithelium with a tall columnar basal layer, subnuclear vacuoles, reverse polarity of hyperchromatic nucleus, and a thin layer of oedematous, degenerating stellate reticulum-like cells on the surface. The term mural UCA is used when the thickened lining (either plexiformor follicular)

penetrates the adjacent capsular tissue.

In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann classified this entity into the following three histologic groups:

Group I—luminal UA (tumor confined to the luminal surface of the cyst);

Group II—intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall);

Group III—mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium). According to this classification, our case study belongs to Group III.

Histologic subgrouping by Philipsen and Reichart has also been described:

Subgroup 1—luminal UA;

Subgroup 1.2—luminal and intraluminal; (our case)

Subgroup 1.2.3—luminal, intraluminal and intramural;

Subgroup 1.3—luminal and intramural.

A definitive diagnosis of unicystic ameloblastoma can only be done by histological examination of the entire lesion and cannot be predicted preoperatively on clinical or radiographic grounds.

The pathogenesis of cystic ameloblastomas remains obscure. Leider et al. (1985) proposed three pathogenic mechanisms for the evolution of UA:

(1) The reduced enamel epithelium which is associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development.

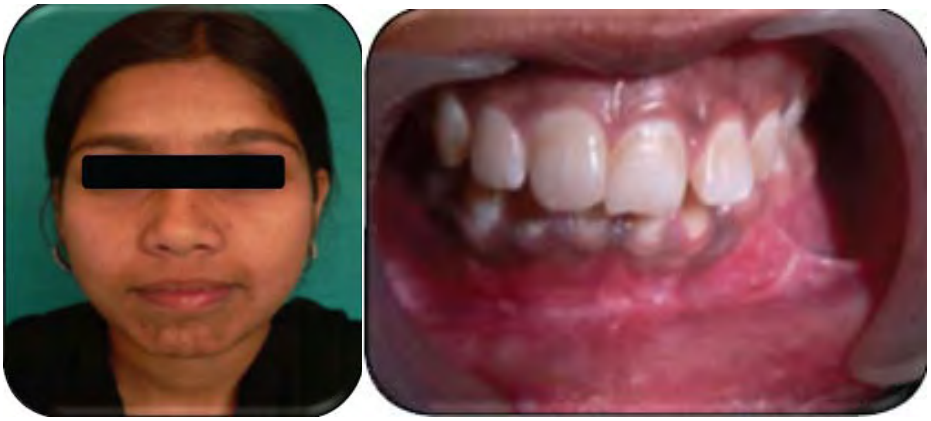
(2) Ameloblastomas arise in dentigerous cysts or in others in which the neoplastic ameloblastic epithelium is preceded temporarily by a nonneoplastic stratified squamous epithelial lining.

(3) A solid ameloblastoma undergoes cystic degeneration of the ameloblastic islands, with subsequent fusion of multiple microcysts and develops into unicystic lesions.

Several attempts have been made in the past to distinguish the lining of the UCAs from that of odontogenic cysts. Eversole et al. contend that currently unaided histologic assessment for UCA remains the gold standard for diagnosis, because of a variable response of UCA to tissue markers.

Treatment planning depends on the histological type of UA. The UA which is diagnosed as subgroups 1 and 1.2 may be treated conservatively (careful enucleation), whereas Subgroups 1.2.3 and 1.3 should be treated aggressively. The recurrence rate for UAs after conservative surgical treatment (curettage or enucleation) is generally reported to be 10–20% and on average, less than 25%. This is considerably less than 50–90% recurrence rates which are noted after the curettage of conventional solid or multicystic ameloblastomas. Lau and Samman reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialization followed by enucleation (where the lesion is reduced in size).

Whatever surgical approach the surgeon decides to take, long-term follow up is mandatory as recurrence of unicystic ameloblastoma may be long delayed. The case was followed for 9 months; there was no recurrence noted till now.



7. Conclusion:

The diagnosis of unicystic ameloblastoma was based on clinical, radiological, histopathologic, and CT features. It is a tumor with a strong propensity of recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. Radiographically, most of ameloblastomas show multilocularity, whereas unicystic ameloblastomas show a single large unilocular radiolucency. Very rarely, we come across a case with presentation of both multilocular and unicystic type in the same person crossing midline. Unicystic variant of ameloblastoma with aggressive histologic behavior also might be successfully treated with marsupialization with subsequent enucleation, and this approach can be considered as an alternative to resection.