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Unicystic Ameloblastoma: Case Reports And Review Of Literature

Case Report

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Introduction

The term unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a jaw cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth [1].

The term Unicystic Ameloblastoma was adopted in the second edition of the international histologic classification of odon togenic tumors. Unicystic ameloblastoma was first described by Robinson and Martinez in 1977 [2]. It accounts for 10-15% of all intraosseous ameloblastomas [3]. The term unicystic ameloblastoma is derived from the macro- and microscopic appearance, the lesion being a well defined, often large monocystic cavity with a lining, focally but rarely entirely composed of odontogenic (ameloblastomatous epithelium). It is often accompanied by an innocuous epithelium of varying histologic appearance that may mimic the lining of a dentigerous or radicular cyst [4].

There can be three pathologic mechanisms for the evolution of UA.

1)The reduced enamel epithelium associated with the developing tooth undergoes ameloblastic change with subsequent cystic transformation.
2)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 lining.
3)Solid ameloblastoma undergoing cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts and then into a unicystic lesion.

With a rich case bank established over 3 decades we have been able to publish extensively in our domain [5-15].

Case Report - I

A 40 year old female patient reported to the department of oral and maxillofacial surgery, Saveetha dental college. The patient reported to the department after developing pain in the lower front tooth region after a fall. On examination swelling was noted on the 41 to 45 tooth region. 43 and 44 were missing clinically.There was no history of pain, toothache, pus discharge or paresthesia. An OPG was taken for radiographic diagnosis.The radiograph reveled a well defined radiolucency from 41 to 45 region .The swelling was not extending to the lower border of the mandible.Biopsy specimen was collected and send for histopathological study .The biopsy report was suggestive of ameloblastoma. Surgical removal of the tumor was planned under general anesthesia. The patient was prepared for the surgery.Crevicular incision was used to raise the mucoperiosetal flap from 41 to 45 region. The tumor was exposed .The tumor was removed intoto. No tooth was removed.The specimen was sent for histopathology .The report reveled it was Type III Unicystic Ameloblastoma (Follicular).

Case Report - II

A 16 year old male patient reported to Oral Maxillofacial surgery at Saveetha dental college with swelling in the lower front tooth region .There was no history of pain, toothache, pus discharge or paresthesia. On intraoral examination,swelling was noted from 42 to 36 tooth region .The swelling was slightly fluctuant.There were no missing tooth.

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Figure 1 a,b,c,d,e,f: (a) Pre operative image of the patient. (b) OPG reveling the well defined radiolucency extending from 41 to 45 region .(c)&(d) intra operative image of exposure and removal of the tumor. (e) closure. (f) Histopthological report suggesting.

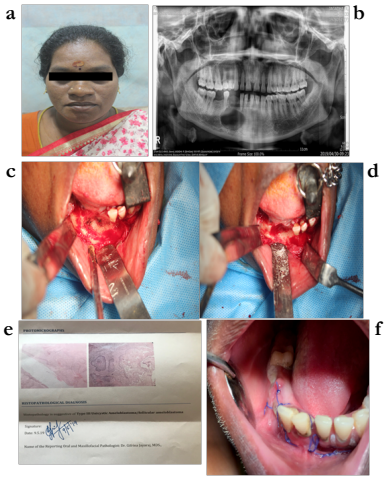
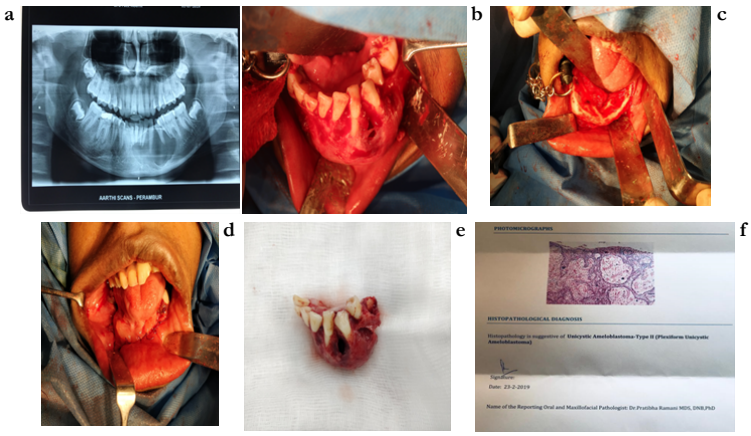


Figure 2 a,b,c,d,e,f: Image (a) is the OPG preoperatively which shows well defined radiolucency from 42 to 46 region.(b) exposed tumor site(c) tumor site after surgical excision of the tumor(d) closure (e)excised tumor specime involving 42 to 35 region.(d) Histopathological report suggestive of Type II Unicystic ameloblastoma(PLEXIFORM TYPE).



On radiographic examination, panoramic view showed a well defined radiolucency extending from distal of 42 to meisal of 46 .Inferiorly the radiolucency was extending 5cm away from lower border of mandible. Aspiration yielded blood. A working diagnosis of ameloblastoma was made on the basis of clinical and radiographic findings.

Incision biopsy showed features of plexiform UA. Even though the patient was young, a conservative treatment was not possible because of extensive involvement. Part of the mandible with the lesion was resected under general anesthesia .Teeth from 44 to 36 were removed and was planned for prosthetic rehabilitation.

The histopathology report was confirmatory of Unicystic ameloblastoma TYPE II (plexiform). The patient is currently unmeder follow up and no recurrence has been reported so far.

Discussion

The ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontoma outnumbers it in reported frequency of occurrence [16]. Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm.

Ameloblastoma, according to the new classification approved at the Editorial and Consensus Conference held in Lyon, France in July 2003 in conjunction with the preparation of the new WHO Blue Book volume Pathology and Genetics of Tumors of the Head and Neck is included under benign neoplasms and tumor-like lesions arising from the odontogenic apparatus showing odontogenic epithelium with mature fibrous stroma, without ectomesenchyme and is divided into four types [4, 16].

1. The classic solid/multicystic ameloblastoma (SMA)
2. The UA
3. The peripheral ameloblastoma (PA)
4. The desmoplastic ameloblastoma (DA), including the so-called hybrid lesions.

Some of the terms used for UA prior to 1977 were cystic ameloblastoma, ameloblastoma associated with dentigerous cyst, cystogenic ameloblastoma, extensive dentigerous cyst with intracystic ameloblastic papilloma, mural ameloblastoma, dentigerous cyst with ameloblastomatous proliferation and ameloblastoma developing in a radicular cyst.

The UA occurs in a younger age group, with slightly more than

50% of cases occurring in patients in the second decade of life. In more than 90% of cases, the UA is located in the mandible, with 77% located in the molar ramus region (mandible to maxilla 13:1) [17]. Between 50 and 80% of cases are associated with tooth impaction, the mandibular third molar being most often involved. The dentigerous type occurs on average 8 years earlier than the non-dentigerous variant. The mean age for unilocular, impaction-associated UAs is 22 years, whereas the mean age for the multilocular lesion unrelated to an impacted tooth is 33 years There are no reports of any sexual or racial predilection [18, 19]. UA dentigerous variant may show a slight male predilection while this ratio is reversed in cases of UA not associated with impacted tooth.

Patients most commonly present with chief complaints of swelling and facial asymmetry. Although the swelling is typically asymptomatic, pain is an occasional presenting sign. A chief complaint of painless swelling often indicates a lesion of long duration and significant size. Continued growth of the tumor and enlargement of the involved area may eventuate in ulceration of the mucosa overlying the lesion. Small lesions tend to be discovered more often on routine radiographic screening examinations or as a result of local effects produced by the tumor. Such local effects include tooth mobility, occlusal alterations and failure of eruption of teeth [20].

Maxillary UAs are very rare. The first case reported by Gardner and colleagues in 1987 occurred in a 12 year old boy in the molar area. There was no bone infiltration. UA in the anterior maxilla is considered to be rare and atypical.

Radiograph of UA presents with unilocular and multilocular patterns with clear predominance for unilocular configuration. Unilocular pattern is often misdiagnosed as an odontogenic keratocyst or Keratinizing Cystic Odontogenic Tumor(KCOT) or a dentigerous cyst and is seen in cases associated with tooth impaction. However, it is stressed that although the lesion is pathomorphologically unicystic, it will far from always produce a unilocular radiolucency. Eversole et al. were able to identify six radiographic patterns for UA ranging from well defined unilocular to multilocular appearances.

Truly multilocular UAs are not encountered often. The scalloping of the cortex and differential bone loss also produces the illusion of a multilocular process on the plane films. The scalloping resorption of the cortical plates rather than compartmentalized areas separated by true bony septa can be visualized in CT images. Contrast-enhanced magnetic resonance imaging was considered useful in the diagnosis of UA, as characteristic features of this type of lesion i.e., thick enhancement of the tumor wall and small intraluminal nodules were detected only by CE-MRI [20, 21].

Histologically, the minimum criterion for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. UA should be differentiated from odontogenic cysts because the former has a higher rate of recurrence than the latter [20, 21]. In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann et al. classified this entity into 3 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).

Histologic subgrouping (modified after Ackermann et al.) by Philipsen and Reichart.[4]

Subgroup 1 - Luminal UA
Subgroup 1.2 - Luminal and intraluminal
Subgroup 1.2.3 - Luminal, intraluminal and intramural Subgroup 1.3 - Luminal and intramural

Plexiform UA, the histologic equivalent of intraluminal UA and coined by Gardne [22] refers to a pattern of epithelial proliferation that has been described in dentigerous cysts. It does not exhibit the histologic criteria for ameloblastoma published by Vickers and Gorlin. Plexiform UAs are not always associated with unerupted teeth, in which case they probably occur over a wider age range than those resembling dentigerous cysts [16]. It exhibits a low rate of recurrence following enucleation or curettage.

The UAs diagnosed as subgroups 1 and 1.2 may be treated conservatively (careful enucleation), whereas subgroups 1.2.3 and 1.3 showing intramural growths must be treated radically, i.e., as a solid or multicystic ameloblastoma [4]. Vigorous curettage of the bone is discouraged since it may implant foci of ameloblastoma more deeply into bone. Chemical cauterization with Carnoy s solution is also advocated for subgroups 1 and 1.2. Subgroups 1.2.3 and 1.3 in which the cystic wall is involved with islands of ameloblastoma tumor cells and there is possible penetration into the surrounding cancellous bone are thought to be associated with a high risk for recurrence, requiring more aggressive surgical procedures [23, 24].

Because the presence of islands of odontogenic epithelium in the cyst wall influences the surgical approach, it is recommended that pathologists carefully examine cystic ameloblastoma surgical specimens for their presence. Multiple, even serial sections are required for examinations. The true nature of these lesions becomes evident only when the entire specimen is submitted for microscopy. The pathology report should include a description of the islands with an indication of their site in the capsule of the tumor. Also treatment plan should take into account factors like individual patient considerations, clinical judgment of the surgeon, type of jaw involved and whether recurrence has occurred or not. Average interval of recurrence is 7 years. Recurrence is also related to histologic subtypes of UA, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7%.

Recurrence rates were 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 reduced in size) or resection [25].

Three cases of UAs are presented with review of literature highlighting histologic variants and mode of treatment.

First case presented, is a subgroup 1 lesion with no infiltration into the surrounding bone. Fortunately, the surgical conduct was

compatible with the biological nature of unicystic ameloblastoma, which does not present an aggressive clinical behavior. In addition, scrupulous review of the surgical specimen revealed the absence of ameloblastic cell chains infiltrating the fibrous capsule, indicating a good prognosis and low recurrence potential. This case illustrates the obvious need for meticulous histologic examination of every cystic lesion of the jaw.

Case two may be treated as a subgroup 1.2 UA. This is the most common presentation of UA. Even though the treatment for this type is theoretically enucleation and curettage, because of of the extensive size of the lesion, resection of affected side of mandible was done.

Despite the fact that UA may, in general, compare favorably with its solid or multicystic counterpart in terms of clinical behavior and response to treatment, the tumors containing invading islands in the fibrous wall could have a high risk of recurrence. The treatment should be in correlation with the histologic and clinical behavior of the lesion. Furthermore, recurrence of UA may be long delayed, and a long-term postoperative follow up is essential to the proper management of these patients. Although the histologic pattern may have implication for the likelihood of recurrence, it should not affect treatment decision. The growth pattern, the jaw in which the tumor is found, age of the patient and histopathologic subtypes are the most important factors when considering treatment options.

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