
The Unicystic Ameloblastoma: A Clinicopathologically Distinct Entity

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Abstract: *Classification of ameloblastomas into solid or multicystic, unicystic, peripheral and desmoplastic types based on the biological behaviour has gained recent recognition in the new WHO classification of head and neck tumours. The distinctive clinical and pathological features of the unicystic variant were reviewed. The variability of histological spectrum, diagnostic problems and choice of treatment for this subtype of ameloblastoma were discussed in the light of recent research interest and progress.*

Key words: *ameloblastoma, odontogenic tumour, unicystic variant*

The ameloblastoma is a benign but locally aggressive neoplasm derived from odontogenic epithelium. It is the most common type of odontogenic tumour, but even so, only accounts for approximately 1% of all oral tumours¹⁻³. Ameloblastomas exhibit a number of histological appearances that can usually be described as follicular, plexiform, acanthomatous, granular cell and basal cell types. However, there appears to be no consistent correlation between these histological patterns and their clinical behaviour^{4,5}, and therefore such microscopic subtyping of ameloblastomas has now essentially become an academic exercise bearing little therapeutic and/or prognostic implications.

The classification, with respect to behaviour, that currently appears to be commonly accepted, separates ameloblastoma into solid or multicystic, unicystic, pe-

ripheral and desmoplastic subtypes, with further separation of these variants based on individual clinical, radiographic and microscopic features^{6,7}. There is increasing justification for regarding these variants as distinct entities and it is no longer appropriate to discuss ameloblastomas as if all cases were essentially similar. In fact, the new WHO classification of head and neck tumours published in 2005 does not simply classify ameloblastoma as a single entity⁸. Rather, it recognises the existence of variants, by using the plural term: ameloblastomas. Four ameloblastoma variants mentioned above are now recognised and the bioprofiles of these ameloblastomas vary in relation to age, distribution, localisation, imaging features, and in particular, prognosis⁸. The unicystic variant of ameloblastoma, for example, has now been stated to be less aggressive than its solid or multicystic counterparts and should be treated by enucleation or curettage⁸⁻¹³. Although considerable insight into the biological profile of unicystic ameloblastoma has been accumulated in recent years, numerous problems with respect to the histogenesis, diagnosis, treatment and prognosis remain obscure. The present paper intends to give a brief overview of various aspects of unicystic ameloblastoma in light of current opinion and development.

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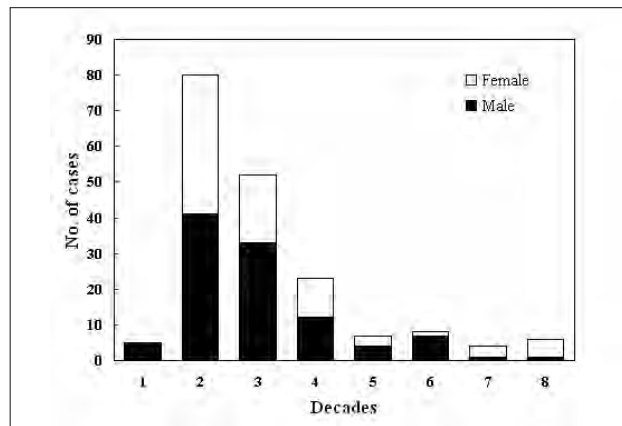


Fig 1 Age and sex distribution of 185 patients with unicystic ameloblastoma (data summarised from references 9,10,12,13,18-33,45).

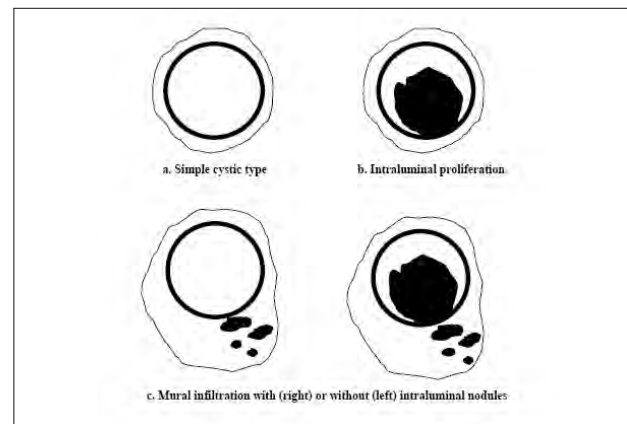


Fig 2 Diagram showing 3 subtypes of unicystic ameloblastoma: (a) simple cystic type; (b) intraluminal proliferation of tumour nodules; (c) infiltration of tumour islands into the cyst wall with or without intraluminal nodules.

Terminology

Unicystic ameloblastoma was first described as a distinct entity by Robinson and Martinez in 1977⁹, although there were several references to this lesion in earlier literature¹⁴⁻¹⁷. It has been termed variously as mural ameloblastoma¹⁸, intracystic ameloblastoma¹⁹, cystogenic ameloblastoma²⁰, cystic ameloblastoma¹⁰, and plexiform unicystic ameloblastoma^{11,21}. The variation of terminology reflects the initial confusion among the authors about the origin of this variant, as it was not clear whether unicystic ameloblastoma originates from a pre-existing odontogenic cyst, i.e. a dentigerous cyst^{10,11,21} or if it forms *de novo*⁹. Since the use of other terms, such as cystic or plexiform unicystic ameloblastoma, tends to fall within the histological sub-groupings previously referred to, it is suggested that the term unicystic ameloblastoma should be uniformly adopted²² and indeed has been quoted in the recent two editions of the WHO classification of odontogenic tumours^{5,8}.

Clinical features

A review of English-language literature from case reports and minor reviews since 1977 disclosed a total number of 215 cases of unicystic ameloblastoma^{9-13,18-34}. There were 185 reported cases with relevant information of the age and sex. The mean age of the patients at the time of diagnosis was 25.4 years, ranging from 6 to 79 years with 44.2% occurring in the second decade (Fig 1). This data indicated that unicystic ameloblastomas tended to occur at an earlier age than the solid or multi-

cystic ameloblastomas. The average age of the later group has been recently reported as 35.9 years with a peak incidence in the third decade³⁵. Possible reasons for this remain obscure but it does lend support for the separation of these two groups. The sex distribution of the present 185 patients [104 male (56.2%); 81 female (43.8%)] were very similar to that reported for solid or multicystic ameloblastoma (male 53%; female 47%)³⁵. Interestingly, unicystic ameloblastoma appears to occur almost exclusively in the mandible. Of the retrieved 164 cases with detailed record of location, 155 (94.5%) affected the mandible, where they had a distinct predilection for the third molar and ramus region (127/155, 81.9%). Unilocular radiolucency with root resorption is a predominant radiographic finding, but a multilocular pattern with well-delineated margins is also discernible in some cases and these tumours may exhibit pseudopod-like cystic extensions grossly and microscopically. Unicystic ameloblastoma is often associated with an unerupted tooth and sometimes assumes a true dentigerous arrangement. Among the 176 reported cases with detailed radiographic description, 63 cases (35.8%) appeared to be related to the crown of an unerupted tooth. This specific feature has led some authors to argue the possibility that unicystic ameloblastoma arises from a pre-existing dentigerous cyst^{10,11}.

Histological features

The classical histological features of unicystic ameloblastoma have been established by several authors^{9,11-13,22}, which all recognised three basic histo-

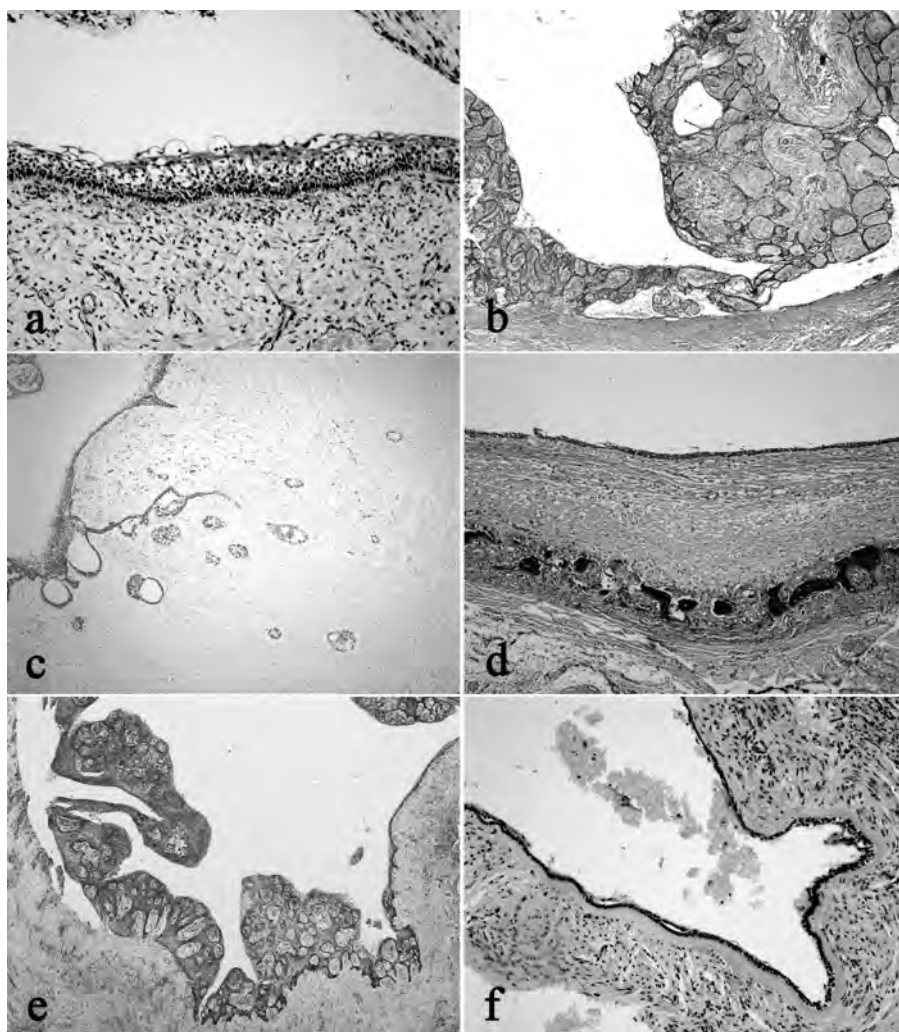


Fig 3 Various histological appearances of unicystic ameloblastoma. The epithelial lining of unicystic ameloblastoma shows, usually in part, typical features of ameloblastoma (A, H-E, x 200). Variants of unicystic ameloblastoma showing predominantly intraluminal plexiform proliferation of the epithelium (B, H-E, x 100) or invasion of tumour islands into the cyst wall (C, H-E, x 100). The cyst cavity may also be lined partially by a non-specific epithelium, such as a thin epithelium of 2–3 cell layers similar to dentigerous cyst lining (D, H-E, x 100), or a thick, hyperplastic epithelium arranged into proliferating arcades, somewhat mimicking radicular cyst linings (E, H-E, ? 40). Occasionally, hyalinization of the connective tissue adjacent to the epithelium is seen (F, H-E, x 150).

logical variants (Fig 2). In the first, a unilocular cystic lesion lined by epithelium which, in some areas, shows the criteria defined by Vickers and Gorlin³⁶ for the diagnosis of ameloblastoma (i.e. columnar basal cells with hyperchromatic nuclei, nuclear palisading with polarisation and cytoplasmic vacuolation with intercellular spacing; Fig 3A). Inactive odontogenic rests might be present within the fibrous wall, but there is no infiltration of neoplastic epithelium. In the second variant the cystic lining is similar to that of the first, but a localised nodule arises from part of this cyst lining and projects into the lumen of the cyst. The intraluminal nodule comprises odontogenic epithelium with a plexiform pattern that closely resembles that seen in the plexiform ameloblastoma (Fig 3B). There is no evidence of tumour infiltration of the fibrous cyst wall. This type is sometimes referred to as the plexiform unicystic ameloblastoma^{11,21}.

Unlike the first two types, the third type of lesion contains tumour islands invading the fibrous wall (Fig 3C). The invading tumour components may show plexiform or follicular pattern of ameloblastoma. Cyst linings partly showing ameloblastomatous features and/or intraluminal tumour nodules may also be present.

Substantial portions of the cyst lining in all three variants may lack the cytological features that are typical of ameloblastoma, and may instead be lined by a non-specific epithelium exhibiting various histological appearances (Fig 3D to 3F). Such cases pose problems in histological diagnosis, and cognisance must be taken of clinical and radiological details. It is essential, therefore, to sample multiple areas from the specimen before making a definitive diagnosis²². Several attempts have been made to distinguish the cystic lining of unicystic ameloblastoma and that of odontogenic cysts using a va-

riety of markers. Gardner et al³⁷ have failed to demonstrate any consistent differences in the expression of blood cell carbohydrates A, B, and H type 2 between odontogenic cysts and unicystic ameloblastomas. However, Saku et al reported that staining for the lectins *Ulex europaeus* agglutinin I (UEA-I) and *Bendeirea simplicifolia* agglutinin I (BSA-I) assisted in the differentiation between cystic ameloblastoma and odontogenic cysts. They found that in the non-neoplastic cysts most of the epithelial linings showed positive binding with UEA-I and BSA-I. No positive reaction was obtained for these two lectins in the solid and the cystic lining components of ameloblastoma, except for limited UEA-I binding to keratinised cells in four cases³⁸. Whilst immunocytochemical localisation of epidermal growth factor receptors (EGF-R) showed no detectable difference between odontogenic cysts and ameloblastomas including the unicystic variant³⁹, epithelial linings of unicystic ameloblastoma contained significantly more Ki67 and PCNA positive cells than that of dentigerous and radicular cysts⁴⁰⁻⁴². Thus, immunocytochemical markers for lectins and proliferating cells may be helpful in differential diagnosis. Li et al¹³ have reported that sub-epithelial hyalinisation within the fibrous tissue wall, a feature rarely seen in odontogenic cysts (except for calcifying odontogenic cysts), is detected in nearly half of their cases (Fig 3F). Given the fact that the submitted biopsy specimens are usually fragmentary in nature and consist of only small portions of the lesion, the presence of this characteristic hyalinised zone in an otherwise non-specific cyst wall should alert the pathologist to the possibility of unicystic ameloblastoma¹³.

Behaviour

Robinson and Martinez⁹ were the first to suggest that the behaviour of unicystic ameloblastoma is less aggressive than the solid types. This was further supported by the subsequent clinicopathological studies and case reports^{10-13,18,19,23-26,28-30,32,34}. The reported rate of recurrence from previous series varied^{3,5,6,12-14,22,24,38}, which may be explained by variations of the follow-up period. As the length of follow-up period for each individual case was not always stated by the authors, it is difficult to compare these data directly. In a large series reported by Li et al¹³, the average interval between initial treatment and obvious recurrence was about 7 years and all recurrences were recorded 4 or more years after initial surgery. Thus, inclusion of cases with follow-up period of less than 4 years could probably result in an underestimation of recurrence rate. In patients with 4 or more years of follow-up, Li et al¹³ reported that a recur-

rence rate of 35%, the highest rate among the previously reported series, was probably a reflection of such a possibility. Even so, this reported figure still compares favourably with the 55% to 90% recurrence rate quoted in various references for ameloblastomas in general (i.e. where solid/unicystic variants are not differentiated) that had been treated by enucleation or curettage^{43,44}.

Although many papers have appeared in the literature indicating a less aggressive nature for unicystic ameloblastoma, few reports are prospective studies examining individual histological variants with respect to behaviour and treatment. The probable reason for a good prognosis is that this variant of tumour is generally well-localised and surrounded by the fibrous capsule. However, once the tumour has breached the periphery of the fibrous tissue wall, it can behave like a solid or multicystic ameloblastoma^{11-13,22}. In five out of six recurrent unicystic ameloblastomas reported by Li et al¹³, the primarily enucleated cystic tumours contained tumour islands in the cyst wall. Thus, unicystic ameloblastomas known to include invasive tumour islands within their cyst walls should indicate a high risk of recurrence. The proliferative activity of unicystic ameloblastoma, as assessed by PCNA and Ki67 labelling indices, has been demonstrated to vary between different epithelial components⁴². There was a progressive increase in the epithelial proliferative activity from cystic tumour lining, through the intraluminal nodules, to the invasive islands in unicystic ameloblastoma⁴². These data further justify the management of lesions exhibiting only cystic and/or intraluminal nodule components by conservative surgical means, whereas lesions containing invasive tumour islands within the cyst walls should indicate an aggressive surgical approach.

Diagnostic problems and choice of treatment

In general, the clinical and radiographic features of unicystic ameloblastoma showed considerable similarities to those of odontogenic jaw cysts. In fact, most of the reported cases were provisionally diagnosed as jaw cysts (odontogenic keratocyst, dentigerous cyst or non-specified cystic lesions) prior to surgery¹³. Interestingly, some authors believed that cases provisionally diagnosed as jaw cysts were more likely to recur in comparison to patients who were initially treated under a clinical diagnosis of ameloblastoma¹³. Whilst this may emphasise the importance of a putative diagnosis based on clinical and radiographic findings, preoperative diagnosis of unicystic ameloblastoma is difficult or sometimes impossible. Since incisional biopsy usually consists of only small fragments of the cyst wall, a positive biopsy

would naturally have rested upon pure chance. The fact that unicystic ameloblastoma could be radiographically multilocular further complicates the problem. From a practical point of view therefore, an expansile unilocular radiolucency, associated with an impacted mandibular third molar or root resorption, occurring in a teenager or young adult should arouse suspicion of unicystic ameloblastoma¹³. Since the definite preoperative diagnosis of unicystic ameloblastoma is often difficult, and sometimes impossible, there appears to be justification that the surgeon should treat any cystic lesions of the jaws as if it contained an ameloblastoma, that is, by careful enucleation. The true nature of the lesion may only become evident when the entire specimen is available for pathological examination. Furthermore, it is crucial for pathologists to carefully examine all biopsy or excision specimens, through multiple sampling or even serial sectioning, to search for ameloblastomatous components, especially to identify the presence of invading tumour islands within the cyst walls^{12,13,32,42}. This not only constitutes the diagnostic procedures but also provides evidence for whether the treatment modality should involve a second operation to remove surrounding bone and/or a long period of follow-up^{12,13,42}.

The age of the patient is another influencing factor relating to the choice of therapy. When dealing with children or teenage patients, an important consideration would be the problem of deformity caused by surgical treatment^{12,17,35}. As unicystic ameloblastoma tends to affect young adolescent patients, the concern to minimise surgical trauma, and to permit jaw function and growth to proceed reasonably unimpaired is one of the important aspects in tumour management¹². Marsupialisation still has a certain amount of respectability despite its obvious limitation of leaving behind cystic tissues. Some authors have reported that marsupialisation can be used as a preliminary treatment for unicystic ameloblastoma^{45,46}, which could help to establish the initial diagnosis and to avoid overtreatment due to faulty diagnosis (i.e. a dentigerous cyst). Some cases have been successfully treated by this method alone⁴⁵ although a second step of surgery is usually required. In any event, long-term follow-up examinations are mandatory in the proper management of patients with unicystic ameloblastoma who have undergone conservative surgery.

Histogenic considerations

Since its first description there has been much debate about whether unicystic ameloblastoma develops de novo^{9,13,22,42} or arises in existing odontogenic cysts, particularly dentigerous cysts^{10,11,21}. In his famous mono-

graph, 'Cysts of the Oral Region', Shear initially described a unicystic ameloblastoma with a dentigerous relationship as 'a dentigerous cyst with mural nodule consisting of tissue very similar to that of a plexiform ameloblastoma'⁴⁷. But the author later reclassified it as 'a unicystic ameloblastoma showing only intraluminal tumour nodules with no infiltration of the cyst wall' in the revised edition⁴⁸. The possibility of an origin from a pre-existing dentigerous cyst is mainly suggested by the fact that unicystic ameloblastomas often involve an unerupted tooth, particularly a third molar at the angle of the mandible. Furthermore, the unicystic lesion may be lined partly by a non-specific, thin epithelium that mimics the dentigerous cyst lining. Thus, provisional radiological examination and biopsy may well regard this type of lesion as consistent with a dentigerous cyst. When the tumour is removed entirely, however, and a diagnosis of unicystic ameloblastoma is made, the lesion may be easily interpreted as having developed from a dentigerous cyst.

On careful examination, however, the involved crown is frequently found to be displaced by the cystic tumour rather than projecting into the lumen as in a dentigerous cyst. Ackermann et al²² reported that only a small percentage of their cases were associated with the crowns of unerupted teeth in a true follicular relationship and there was no evidence that any other odontogenic cyst existed prior to the development of the lesions. Indeed, even a lesion with a true dentigerous relationship does not denote origin of that lesion from the follicle, since an erupting tooth could grow into an adjacent cystic lesion⁴⁹. For example, the so called 'follicular keratocyst', a term referring to an odontogenic keratocyst with a true dentigerous relationship to the associated unerupted tooth, is believed to be an originally extrafollicular lesion, but subsequently, its epithelial lining fuses with the reduced enamel epithelium when the associated tooth erupts into the cyst lumen⁵⁰. Thus, it can be argued that the unicystic ameloblastoma is a variant of the neoplasm in which early cyst formation is a characteristic feature, without postulating the previous existence of a dentigerous cyst. By quantitative immunocytochemistry, Li et al⁴⁰⁻⁴² demonstrated that the proliferative activity of cystic tumour linings in unicystic ameloblastoma is significantly different from those of odontogenic keratocysts, dentigerous and radicular cysts. All areas of cystic tumour epithelium contained significantly more proliferating cells than dentigerous and radicular cyst linings, even in areas where epithelial morphology was similar to dentigerous cyst lining⁴⁰⁻⁴², favouring the concept that unicystic ameloblastoma is a de novo cystic neoplasm.

Summary

The new WHO classification defines unicystic ameloblastoma as an ameloblastoma variant presenting as a cyst⁸. It refers to those cystic lesions that show clinical and radiological characteristics of an odontogenic cyst, but in histological examination show a typical ameloblastomatous epithelium lining part or most of the cyst cavity, with either luminal or mural tumour proliferation¹³. The present review clearly indicates that this group of lesions tends to occur at a younger age than conventional ameloblastoma, and the response to enucleation or curettage is more favourable. Conservative surgery seems to be justified in preference to mutilating radical surgery, despite the obvious risk of recurrence, probably related to the presence of infiltrative tumour components in the cystic wall. In addition to the continued clinical documentation, with particular reference to the long-term follow-up, basic research with respect to elucidate the growth and spreading pattern of unicystic ameloblastoma is of critical importance. From greater understanding of the nature of this tumour variant, the various treatment protocols currently recommended by different surgeons will be better rationalised.

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