

Unicystic Ameloblastoma arising from a Dentigerous Cyst: A Case Report

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ABSTRACT

The lining of an odontogenic cyst may show potential for neoplastic transformation to non odontogenic malignancies like squamous cell carcinoma and mucoepidermoid carcinoma and also odontogenic tumors like ameloblastoma and adenoamatoid odontogenic tumor (AOT). Unicystic ameloblastoma is a single cystic cavity lesion which shows ameloblastomatous differentiation in the lining. The clinical and radiological presentation of UCA can present a confusing picture of an odontogenic cyst, especially when it is seen in the inter-radicular or periapical area. Also, tooth associated UA may show features similar to dentigerous cyst. Hence histopathological examination is essential to diagnose such cases. Here, we present, a case of unicystic ameloblastoma arising from a dentigerous cyst associated with an impacted mandibular third molar. A literature review on the topic has also been added.

KEYWORDS: Dentigerous cyst, Mandible, Unicystic ameloblastoma, unilocular

INTRODUCTION

Odontogenic cysts and tumors represent a surprisingly diverse group of pathologic lesions of the jaws. A basic understanding of the histology and embryology of tooth formation can help in understanding the development and histopathology of these lesions. Non odontogenic malignancies such as mucoepidermoid carcinoma, squamous cell carcinoma and odontogenic tumors like ameloblastoma and adenoamatoid odontogenic tumor (AOT) are known to arise from lining of odontogenic cyst.¹ However; very few such cases have been reported in literature.

In this report, a case of unicystic ameloblastoma arising from a dentigerous cyst in a young female patient is presented.

CASE REPORT

A 20 year old female patient with non contributory medical history reported with complaint of malaligned teeth, for that we advised her radiograph for further evaluation. A panoramic radiograph showed a well defined single unilocular radiolucency associated with unerupted 38 (Figure 1, 2).

Based on clinical and radiological pictures provisional diagnosis of dentigerous cyst was made, and excisional biopsy was done. The cyst was enucleated under general anesthesia and 37, 38 were extracted. There were no post-operative complications and the wound healed uneventfully. On gross examination, the specimen, which was 3x3x2 cm in diameter, consisted of an irregular solid mass of tissue attached to the cemento - enamel junction of 38.

Microscopically, the section showed a cystic lumen lined

partly by thin non keratinized stratified squamous epithelium proliferating in areas with stellate reticulum like changes in the superficial layers. The connective tissue was loose fibro-cellular with thin collagen fibers and moderate chronic inflammatory cell infiltrate. It also showed the presence of epithelial rests (Figure 3).

These findings were suggestive of unicystic ameloblastoma arising from the lining of a dentigerous cyst.



Figure 1. Intraoral image



Figure 2. A panoramic radiograph showing a well defined single unilocular radiolucency associated with unerupted 38

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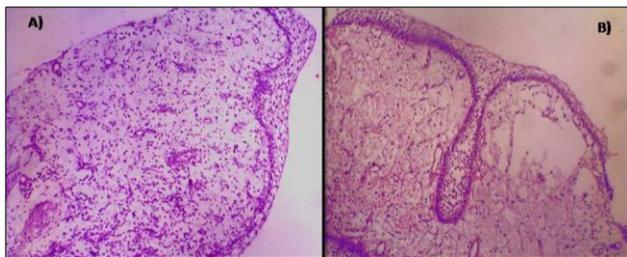


Figure 3 A) and B): Proliferating ameloblastomatous epithelium with stellate reticulum like changes in the superficial layers

DISCUSSION

Dentigerous cyst (DC) is a developmental odontogenic cyst that encloses the crown of an unerupted tooth by expansion of its follicle, and is attached to the neck of the tooth.¹ A dentigerous cyst is the most common cause of pericoronal radiolucency which is associated with impacted teeth.¹⁻³ The diagnosis of a DC is straight forward; but some can be miss diagnosed as a dental follicle, a hyperplastic dental follicle, a keratocystic odontogenic tumour (KCOT) or a unicystic ameloblastoma. The histological diagnosis of these lesions are therefore critical.² Dentigerous cyst is often seen in the age group of 10–30 years and most commonly associated with an unerupted mandibular 3rd molar as seen in our case. They are encountered in almost 1% of individuals having dental roentgenograms.³

The epithelium of odontogenic cysts may be transformed into odontogenic tumors like ameloblastoma and adenomatoid odontogenic tumors, or non odontogenic tumors. Many etiological factors have been proposed for the ameloblastomas arising from odontogenic cysts, including; nonspecific irritational factors (extraction, trauma, infection, inflammation, unerupted tooth), nutritional deficiency, viral infections.³ Ameloblastoma is the most common odontogenic tumor, accounting for 10% of all such tumors. It is a slow-growing neoplasm, usually occurring in young adults 20–40 years old, with almost equal distribution among men and women.⁴ It occurs in the mandible in 80% of the patients and 70% of these arise from the molar-ramus area.³

Unicystic ameloblastomas were first described by Robinson and Martinez. These are cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but on histological examination, they show characteristic ameloblastomatous epithelium lining, with or without a luminal or mural tumour proliferation.⁵ Ackerson, on his clinico-pathological study done on 57 cases of unicystic ameloblastomas, classified this entity into three histological groups: luminal unicystic ameloblastoma, intraluminal/plexiform unicystic ameloblastoma and mural unicystic ameloblastoma.² Almost, 15% to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts. The epithelial lining of the lumen exhibits uniform thickness and hyperchromatic layer of palisaded basal cells, most of which exhibit reversed polarization of the nucleus. The suprabasal layers resemble stellate reticulum.⁶ This

histologic pattern referred to as intraluminal UCA that contains areas in which the epithelium is thickened with papillary projections extending into the lumen. In some of lesions, the luminal projections demonstrate an edematous, plexiform pattern that resembling plexiform pattern of conventional ameloblastomas, and therefore, these lesions are referred to as plexiform cystic ameloblastomas. When the thickened lining penetrates the adjacent capsular tissue, it is termed a mural UCA that may show either plexiform or follicular pattern.^{6,7} A. Seintou et al., reviewed the clinical, radiological, and histopathological profile of unicystic ameloblastomas (UA) in children using data from available case reports and reviews published between 1992 and 2012. The result of this study is shown in Table no.1. These results are consistent with our case such as mandible location and female with age of 20 years.⁸

Today an important controversy regarding this tumor is the differential diagnosis between Ameloblastoma showing cystic degeneration and Ameloblastoma arising from an odontogenic cyst. Many authors suggest that most ameloblastomas arise from dentigerous cysts and the likely explanations for the pathogenesis are the ameloblastic transformation of the cyst lining. According to these authors, the epithelium of the cyst, which surrounds the unerupted tooth and is continuous with the cystic or solid ameloblastoma areas, (like in our case) is originated from the cement-enamel junction of a tooth, and it shows dentigerous relationship with the unerupted tooth. In most cases of odontogenic cysts, the presence of an ameloblastomatous epithelial lining is insufficient to diagnose unicystic ameloblastomas, unless other more diagnostic features of unicystic ameloblastomas are evident such as features described by Vickers and Gorlin such as tall columnar basal layer, reverse polarity of hyper chromatic nucleus, a sub nuclear vacuole, and a thin layer of edematous, degenerating stellate reticulum like cells on surface.

Dentigerous cysts, residual cyst, odontogenic keratocyst, adenomatoid odontogenic tumour, giant cell lesions and sometimes solid Ameloblastoma are possible differential diagnoses for a unilocular lesion with or without a ‘dentigerous’ relationship occurring within the jaws. Residual cysts are associated with missing teeth that have been extracted. Keratocyte usually spread anterior-posterioly and seldom shows cortical expansion. Adenomatoid odontogenic tumors have a predilection for anterior maxilla. Solid ameloblastoma is multilocular, and it is uncommonly seen in patients less than 30 years of age.⁹ Conventionally described treatment is based on exact histology of the UCA.⁶ In cases of the intraluminal or plexiform pattern being present (and no penetration of the fibrous tissue capsule by ameloblastic cells), enucleation generally suffices, but if there is lesional component that extends into the wall to the level of the interface with the bone, bony resection is necessary to ensure adequate removal.⁶

In summary, the present case is consistent with the recognized association of dentigerous cysts and

Male to female ratio	Location	Clinical appearance	Radiographical appearance	Histological type	Treatment	Recurrence
0.96:1	Maxilla: 13.7% Mandible: 86.3%	Painless swelling: 37.2% Routine orthodontic evaluation: 5.9% Mild fullness over the cheek: 3.9% Pain: 3.9% No information: 43.1%	Unilocular: 70.6 % Multilocular: 5.9% No information: 21.6 %	Luminal: 25.5% Intraluminal: 5.9% Mural: 7.8% No information: 60.8%	Enucleation–curettage: 60.8 % Margin resection: 5.9% Decompression before enucleation: 2.0% Excision before enucleation: 2.0 % Marsupialization before enucleation: 3.9% Enucleation + Carnoy’s solution: 5.9%	No recurrence: 60.8% With recurrence: 29.4% (All cases treated with enucleation or excision) No information: 7.8% Lost to follow -up: 2.0%

Table no.1: Overview of the results in percentages and ratios

ameloblastomas in unerupted third molar teeth. The coexistence of these three entities also support the hypothesis of ameloblastic transformation of dentigerous cysts. It is important to remember that the diagnosis of an odontogenic lesion relies heavily on histopathologic evaluation.

CONCLUSION

Preoperative diagnosis of UA can be difficult because this type of ameloblastoma shares significant clinical and radiographic similarities with odontogenic cysts. Thus, a histological examination is considered to be the most sensitive tool which can be used for differentiation of dentigerous cysts from unicystic ameloblastomas. While ameloblastomas, being of odontogenic epithelial origin, may theoretically arise from dentigerous cyst lining as well as any other odontogenic epithelium, the belief that it commonly arises in this situation and that the dentigerous cyst should, therefore, be regarded as pre ameloblastomatous, should be viewed with caution.

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