

Dentigerous Cyst Converting to Ameloblastoma in Anterior Mandible: An Unusual Case Report

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ABSTRACT

Background: A Dentigerous cyst is the most frequent odontogenic cyst of developmental origin mostly associated with impacted mandibular third molars. Very rarely ameloblastoma arises from the epithelium of dentigerous cyst. In this paper we report a case of a dentigerous cyst converting to ameloblastoma in its unusual site i.e. anterior mandible, crossing the midline. Enucleation of the cystic lesion was done under conscious sedation.

Keywords: Ameloblastoma, Dentigerous cyst, mandible.

INTRODUCTION

Odontogenic cysts and tumors are lesions of the oral and maxillofacial region that must be considered whenever examining and formulating a differential diagnosis of expansile pathologies of the jaws.

The clinical presentation, radiographic appearance, and natural history of these lesions vary considerably, such that they represent a diverse group of lesions of the jaws and overlying soft tissues. A dentigerous cyst surrounds the crown of an unerupted tooth, mostly the maxillary canine or the mandibular third molar teeth are quite common.¹⁻² In the mandibular anterior region, occurrence of Dentigerous cyst is very uncommon.

In addition to that pathologic conversion of dentigerous cyst into ameloblastoma is even unusual. We report a case of dentigerous cyst converting to ameloblastoma in anterior mandibular region associated with impacted mandibular canine.

CASE REPORT

A 60 year old female was referred to the Dept of Oral and Maxillofacial Surgery with mild pain in lower front region. (Fig-1) She was treated for pulmonary TB 16 years back and she has undergone extraction of her lower front teeth 5 year before. She reported to the department with pain in lower left tooth region which aggravates on eating. Intraoral examination reveals a diffuse solitary swelling which is causing lower anterior cortical plate expansion from 43 to 35 region. Labial vestibule was obliterated due to swelling. Swelling was of 6 X 3cm of size and it had smooth surface & erythematous surrounding mucosa. [Fig-2]. On palpation it was bony hard with fluctuant surface on superior aspect. It was nontender and non pulsatile. Expansion of buccal cortical plate in relation to 42 – 35 region. Egg shell crackling was felt on palpation of the swelling. Aspiration revealed 1 ml of thin light yellow straw colored fluid. A panoramic radiograph [Fig-3] revealed a single, unilateral, well defined, radiolucent area in the anterior mandible, crossing midline approx 4x2.5 cm, extending from root of 35

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Fig 1: Frontal profile.



Fig 2: Intraoral view.



Fig 3: OPG radiograph.

(mesial aspect) to 1.5 cm medial to 44 Root resorption was present in relation to 34 anteroposteriorly and from lower border of mandible to alveolar crest superoinferiorly. Lower border is intact without any expansion. Submental and submandibular lymphnodes were palpable,



Fig 4: Cross sectional mandibular occlusal view.



Fig 5: Intraoperative photographs.

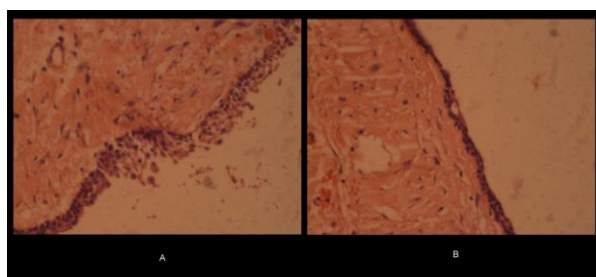


Fig 6: Histopathological examination showing features of Dentigerous cyst (A) and ameloblastoma (B) (Hematoxylin - Eosin x 100).

nontender and mobile. Mandibular cross sectional occlusal radiograph showed well defined 2.5 x 4 cm sized unilocular radiolucent lesion with the epicenter around the mandibular canine. Hydraulic expansion of outer cortical plate was evident without any perforation. (Fig-4) Lingual cortex was intact. Preoperative evaluation was done and complete blood investigations were done. Under

conscious sedation, enucleation of the cystic lesion, along with the extraction of unerupted mandibular canine was done. (Fig-5) On histopathological examination, it was identified that cystic wall was lined by thin stratified epithelium made up of low cuboidal cells. The underlying connective tissue was densely fibrous. The epithelium showed transformation into odontogenic epithelium with loosely arranged stellate cells. (Fig-6) Final histopathological diagnosis was Dentigerous cyst transforming into Ameloblastoma (Unicystic). Sutures were removed after 7 days and healing was uneventful.

DISCUSSION

A dentigerous cyst is the second most common cyst after radicular cyst occurs in association with an unerupted tooth, most commonly mandibular third molars. Other common associations are with maxillary third molars, maxillary canines, and mandibular second premolars.³ In our case it was associated with mandibular impacted canine which is a very uncommon site for Dentigerous cyst. Common lesions encountered in anterior mandible are Central giant cell granuloma, lateral periodontal cyst, Glandular odontogenic cyst etc. Many Dentigerous cysts are small asymptomatic lesions that are discovered serendipitously on routine radiographs, although some may grow to considerable size causing bony expansion that is usually painless until secondary infection occurs.¹ The epithelium of the odontogenic cyst may transformed in to odontogenic tumors like ameloblastoma, Adenomatoid odontogenic tumor etc. The ameloblastoma is the most common clinically important and potentially aggressive odontogenic tumor arise from rests of the dental lamina, a developing enamel organ, the epithelial lining of an odontogenic cyst, or the basal cells of the oral mucosa.⁴ They behave aggressively locally, but do not metastasize. Unicystic Ameloblastoma is a rare variant (6%), which refers to those cystic lesions, that show clinical, radiographic or gross features of a jaw cyst but on histological examination shows a typical ameloblastomatous epithelium.⁵ Approximately 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts.

The development of ameloblastomas within the walls of odontogenic cysts had been reported in the literature by many authors among which the most common entity was dentigerous cysts.⁶⁻⁷ The expression of immune-histochemical marker Ki-67 in ameloblastomas which transforms from dentigerous cysts prove the hypothesis that ameloblastomas which originate from dentigerous cysts have a biological behavior which is similar to that of unicystic ameloblastomas⁸. In view of the reported ameloblastomatous potential of dentigerous cysts, it is important to be able to recognize true ameloblastomatous epithelium from ameloblastoma -like epithelium.⁹ so, the point of debate is differential diagnosis between ameloblastoma showing cystic degeneration and ameloblastomas arising from an odontogenic cyst or the ameloblastomatous hyperplasia in the odontogenic cysts. Authors suggest that most of the ameloblastomas may arise from the dentigerous cysts and the most likely explanation for the pathogenesis is the ameloblastic transformation of the ordinary dentigerous cyst lining. According to this, the epithelium of the cyst, which surrounds the unerupted tooth and continues with the cystic areas, like in our case it shows dentigerous relationship with the unerupted tooth.

A different opinion exists by stating that the embryological origin of the epithelium of both odontogenic cysts and ameloblastomas are similar; thus such kind of relation is usual. According to Shear, much of the confusion has arisen from three reasons: First, an ameloblastoma may involve an unerupted tooth, like dentigerous cysts; and the histopathological evaluation of this specimen may reveal a conclusion that the ameloblastoma is developed from a dentigerous cyst.⁹ Secondly, when the biopsy is taken from a site of an expanded locule lined by a thin layer of epithelium, the histopathological diagnosis may be an ameloblastoma arising from a dentigerous cyst. The third reason is that apparently isolated islets or follicles of epithelium are sometimes found in the cyst wall at some distance from the epithelial lining and they may be interpreted as ameloblastoma, although they bear only a superficial resemblance of the tumor³ which are valid for the presented case. In the present case, the occurrence of dentigerous relation between the simple epithelium of the cystic

cavity has suggested a diagnosis of unicystic ameloblastoma arising from a dentigerous cyst.

CONCLUSION

Odontogenic cyst & tumors of the jaws arising from the tooth forming tissues are usual. Although ameloblastoma is the most common of the epithelial odontogenic tumors, occurrence of unicystic ameloblastoma is still rare and whether the ameloblastoma arises from the dentigerous cysts or not is still controversial. The final diagnosis of benign cyst and tumor is solely based on a histopathological examination. The large lesion may be planned for decompression and marsupialization initially but if the lesion has ameloblastomatous epithelium then it highlights the importance of enucleation as the first choice of treatment.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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