



Ameloblastic Transformation of a Primarily Diagnosed Dentigerous Cyst of Jaw- A Rare Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

The separation of the follicle from around the crown of an unerupted tooth causes formation of dentigerous cyst. It may undergo metaplasia in several cases. In this case a 30 year old female patient reported with a history of dentigerous cyst with respect to impacted 48. The swelling started growing gradually causing facial asymmetry. On intraoral examination the swelling seem to extend from 48 to 46 region and cortical plates were expanded. The histopathological examination revealed the sample to be cystic ameloblastoma (mural variety). This paper sends a message that the potential of a cyst should never be understated, instead it must be kept in an observation or understanding the biological behavior over a course of time.

Keywords: *Ameloblastoma; dentigerous cyst; unilocular radiolucency; cortical plate involvement; odontogenic metaplasia.*

1. INTRODUCTION

The dentigerous cyst (DC) is defined as a cyst that originates by the separation of the follicle from around the crown of an unerupted tooth. It is usually docile but may undergo metaplasia [1]. DC is formed by the accumulation of fluid between the reduced enamel epithelium and the unerupted tooth crown. It is usually associated with unerupted mandibular third molar, maxillary canine, and mandibular premolars [2].

The neoplasms that might develop from the cystic lining of a dentigerous cyst are ameloblastoma and adenomatoid odontogenic tumor [3]. In the year 1933 Cahn first reported such a case where ameloblastoma is developed from an existing dentigerous cyst [4]. Vickers and Gorlin put forth criteria to delineate the transformation of cystic lining into the neoplasm histopathologically [5]. Reported cases of ameloblastoma arising from dentigerous cyst are quite less.

2. CASE REPORT

A 30-year-old female patient reported to outpatient department with mild tenderness and asymptomatic swelling on lower right back tooth region. According to the history given by the patient it was evident that there is a history of impacted 48 which was extracted along with periapical curettage few years back and histopathological reports revealed 2-4 layers of non keratinizing stratified squamous epithelium. Usually epithelium connective tissue interface is flat. Fibrous connective tissue wall is loosely arranged, often as islands and cords of inactive odontogenic epithelial rests seen with diffuse chronic inflammatory infiltrate which was corroborative to dentigerous cyst (DC) (Fig. 1). Initially the swelling was small but later on it increased in size gradually. Extraoral examination revealed facial asymmetry and non tender, hard swelling over the right ramus and border of mandible. Right submandibular lymph nodes were palpable and enlarged.

Intraoral examination revealed the swelling to be extended from 46 to 48 regions and covered by normal mucosa. Both of the buccal and lingual cortical plates were deformed due to the lesion. Missing teeth of the quadrants are 46 47 48. Based on all the previous history and findings a provisional diagnosis of recurrence of DC was given.

As further investigation orthopantomogram (OPG) and cone beam computed tomography (CBCT) was done. The radiographic findings depicts buccal cortical plate involvement of the unilocular lesion and inferiorly extension was upto the inferior alveolar nerve with minimal involvement.(Fig. 2) A three dimensional reconstruction was done along with the CBCT to rule the extension of the lesion in accordance with the nerve. (Fig. 3).

Entire lesion along with the normal tissue was removed surgically from right lower 1st molar to 3rd molar area under general anesthesia. Complete excised specimen was sent to laboratory for histopathologic examination. Bits of specimens were collected in which the entire lining of the suspected cystic lesion were present.(Fig. 4)

Histopathology of the specimen revealed thin cystic epithelium with prominent, palisading basal cells and superficial cells resembling stellate reticulum cells. The fibrous capsule shows multiple follicles and interconnecting strands of odontogenic epithelium with peripheral ameloblast like cells and central stellate reticulum like cells. Moderate infiltration of chronic inflammatory cells in the collagenous tissue stroma. The overall histopathologic features are suggestive of the lesion to be cystic ameloblastoma (mural variant).(Fig. 5)

3. DISCUSSION

Dentigerous cyst formed by accumulation of fluid between reduced enamel epithelium layer and crowns. This dentigerous cyst is usually associated with impacted tooth and accidentally discovered during routine radiographic examination. The follicle seemed to be expanded here. Histopathological picture shows 2-4 layers of flat non keratinizing stratified squamous epithelium. Usually epithelium connective tissue interface is flat. Fibrous connective tissue wall is loosely arranged often islands and cords of inactive odontogenic epithelial rests seen. Inflamed one presents with diffuse chronic inflammatory infiltrate Few literatures described a neoplastic potential of epithelium of dentigerous cyst to transform into ameloblast, epidermoid carcinoma, mucoepidermoid carcinoma etc [3-4].

On the other hand Ameloblastoma can be described as locally aggressive benign epithelial odontogenic tumor. Ameloblastoma can develop from remnants of dental lamina, enamel organ,

basal layer of oral epithelium, and epithelium of DC etc W.H.O 2017 classification divided Ameloblastomas into four categories conventional, extraosseous/peripheral, unicystic and metastasizing ameloblastoma [5].

Histopathologically, it can be divided into: follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic type. According to Vicker and Gorlin, cuboidal or columnar cells with hyperchromatic nuclei, cytoplasmic vacuolization, nuclear palisading, polarization, subepithelial hyalinization etc features indicate a transformation towards ameloblastoma. Shear

described few reasons regarding the relation of dentigerous cyst and its transformation to ameloblastoma. They both share quite a common clinical and radiographic feature, so probability of development of ameloblastoma from dentigerous cyst always exists. The second reason described the biopsy specimen taken from expanded locule can be ameloblastoma developing in epithelial lining of dentigerous cyst. Islands and follicles of ameloblastomatous epithelium can be seen in the wall of dentigerous cyst and this further proliferates to form ameloblastoma [6].

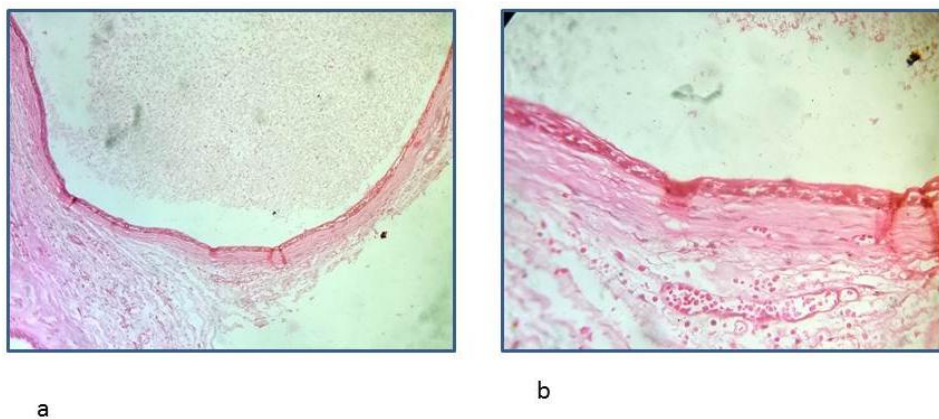


Fig. 1. The initial histopathological presentation of the lesion with features of dentigerous cyst
a. Scanner view – 2-4 cells thick layer with flattened basement membrane, the underlying connective tissue shows fibrous capsule with minimal inflammation and moderate vascularity. B. High power view of the flattened epithelium and fibrous capsule

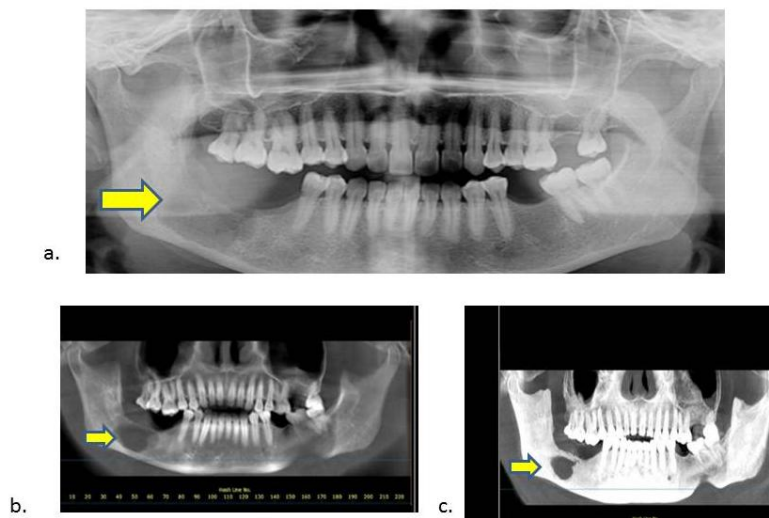


Fig. 2. a. Orthopantomograph showed 46,47,48 appear to be missing. In the edentulous and adjoining ascending ramus area, a bone defect is seen. Inferiorly, the defect extends beyond mandibular canal, falling short of the lower border of mandible. b,c- Endoscan (CBCT) showed presence of a unilocular radiolucency on the region of 46,47,48

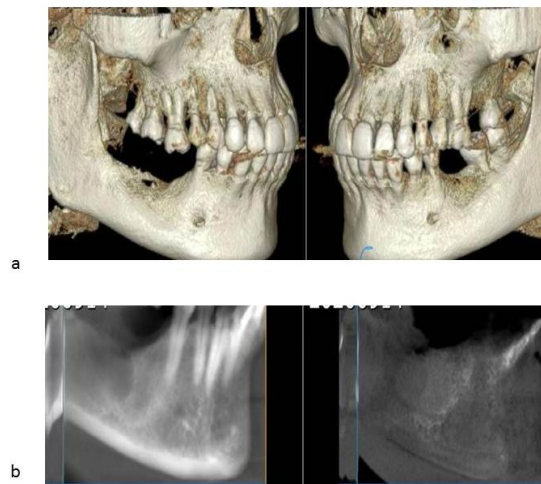


Fig. 3. a.3D reconstruction revealed involvement of buccal cortical plate. B. CBCT revealed the defect extends upto the inferior alveolar canal

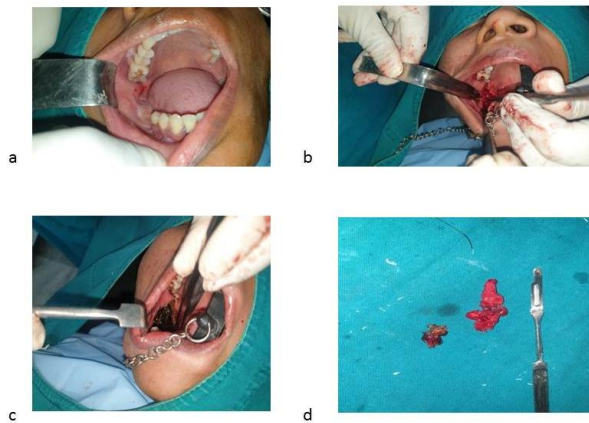


Fig. 4. A. Intraoral swelling extended from 46 to 48 regions and covered by normal mucosa. Missing 46,47,48 noted. b. Pre- operative procedure under G.A c. Application of Carnoy's solution on the surgical area d. Complete excised mass

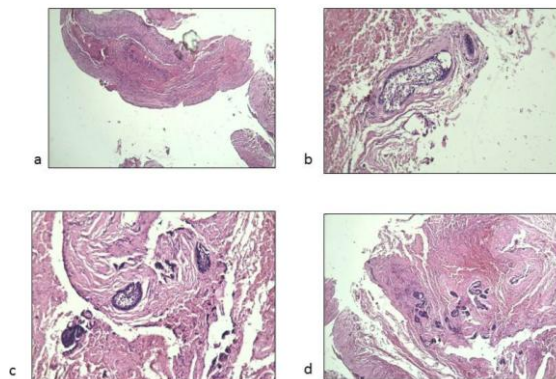


Fig. 5. A. Scanner view of the tumour component entrapped within the dense fibrocollagenous connective tissue b,c- islands and nests of ameloblastic follicles with hyperchromatic odontogenic cells with reversal of polarity and centrally places loose stellate reticulum like cells (satisfying Vickers and Gorlins criteria) d. fibrous component of the tumour

4. CONCLUSION

A rare case of development of ameloblastoma from dentigerous cyst has been described here. Neoplastic potential of dentigerous cyst to transform into ameloblastoma has been described under the light of histopathology in this article. Minute detail of case history, observation and correlation with histopathologic features are important.

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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