

Ameloblastomatous Transformation of Dentigerous Cyst-Report of Two Rare Cases

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Abstract

Dentigerous cyst is a developmental odontogenic cyst that encloses the crown of an unerupted tooth. Dentigerous cyst linings were found to have the potential to transform into ameloblastoma. Two distinct examples of ameloblastoma developing in the wall of a dentigerous cyst are described here.

Keywords: Dentigerous cyst, Ameloblastomatous transformation.

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INTRODUCTION

Dentigerous cyst is the most common developmental cyst in the oral cavity that encloses the crown of an unerupted tooth by expansion of its follicle [1]. The epithelium of dentigerous cysts may be transformed into odontogenic tumors like ameloblastoma or non-odontogenic tumors. Ameloblastoma is the most common odontogenic tumor, accounting for 10% of all such tumors [2].

Here, we present two cases of ameloblastic transformation of dentigerous cyst with different clinicopathological features.

CASE REPORT-1

A 17 year old male patient reported to the department with swelling on the left side of face since 2 months. Extra oral examination revealed a diffuse swelling of size 2 x 1.5 cm extending 1.5 cm away from the left corner of lip to the posterior body of mandible. The swelling was firm to hard in consistency and tender on palpation. Single left submandibular lymph node of size 0.5 x 0.5 cms in size was palpable, which was firm in consistency, mobile and tender.

On intra oral examination, obliteration of buccal vestibule and expansion of the buccal cortical plate were noted in relation to 36 to 38 region 36 and 37 was tender on percussion (Figure 1).



Figure 1: Expansion of buccal cortical plate in relation to 37 and 38 region

Orthopantomograph revealed well defined radiolucency of size 2.5 x 2 cm with sclerotic border extending from the mesial root of 37 to mesial aspect of 38 and horizontally impacted 38 (Figure 2). The inferior alveolar canal was inferiorly displaced with intact lower border of mandible. Cone-beam computed tomography revealed buccal and lingual cortical expansion with cortical perforation at 37 regions (Figure 3).



Figure 2: Well defined radiolucency of size 2.5 x 2 cm with sclerotic border extending from the mesial root of 37 to mesial aspect of 38 and horizontally impacted 38

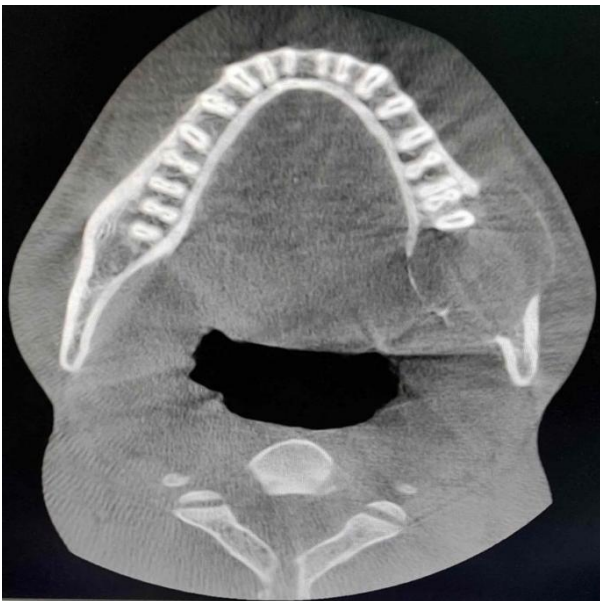


Figure 3: Buccal and lingual cortical expansion with cortical perforation at 37 regions

Based on the clinical and radiological features, a provisional diagnosis of Dentigerous Cyst (DC) was given. Marsupialization of the cystic lesion was done after extraction of 38 under local anaesthesia. Cystic lining enucleated from the distal aspect of 37 was sent for histopathological examination.

Histopathological examination revealed cystic lumen lined by non-keratinized epithelium of 2-3 cell layers thickness (Figure 4) and in a few areas odontogenic epithelium proliferating into the lumen was noticed (Figure 5). Suprabasal layer of cystic lining showed stellate reticulum like cells in a few areas. The underlying connective tissue stroma shows ameloblastic islands with reversal of polarity of nuclei and cytoplasmic vacuolizations (Figure 6). The histopathologic findings were similar to those of an ameloblastoma developing within the lining of a dentigerous cyst. Patient was referred to the department of oral and maxillofacial surgery for further treatment.

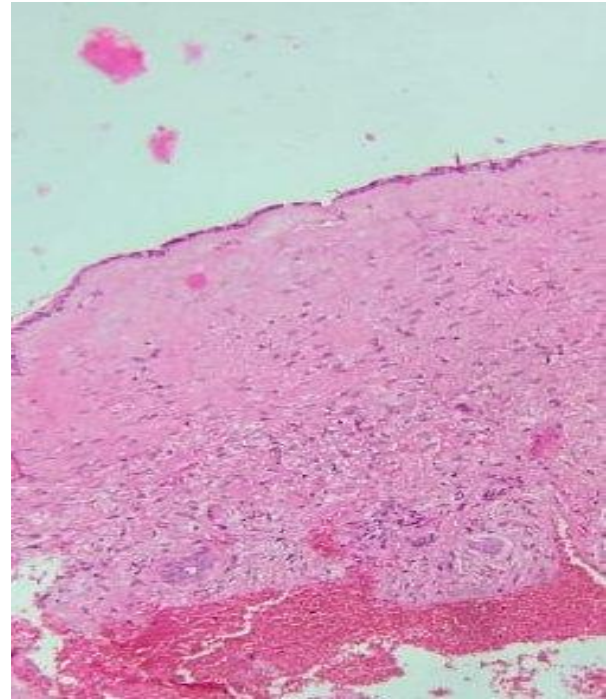


Figure 4: Dentigerous cyst lining (H & E, 4x)

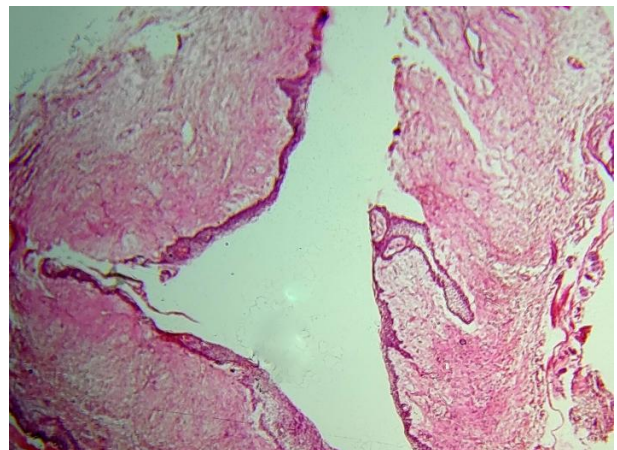


Figure 5: Odontogenic epithelium proliferating into the lumen (H & E, 10x)

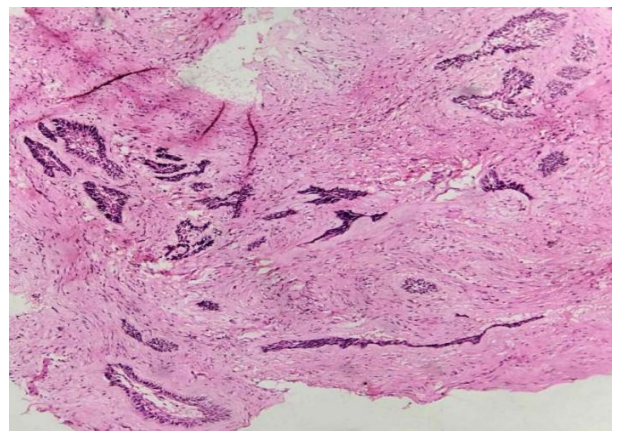


Figure 6: Ameloblastic foci (H&E, 10x)

CASE REPORT -2

A 25-year-old male patient reported with a chief complaint of intermittent pain in the left cheek region for the past two weeks. The patient had a history of dentigerous cyst in mandibular left third molar region 4 years back which was treated with enucleation and surgical extraction of the impacted tooth. Orthopantomograph of the previous lesion revealed well defined radiolucency involving impacted 38 and extending posteriorly to mandibular ramus upto sigmoid notch. It was well corticated with a scalloped border (Figure 7). Biopsy showed 2 to 4 cell layers thick non-keratinized epithelium overlying myxomatous connective tissue stroma (Figure 8).



Figure 7: Orthopantomograph revealing well defined radiolucency involving impacted 38 and extending posteriorly to mandibular ramus upto sigmoid notch

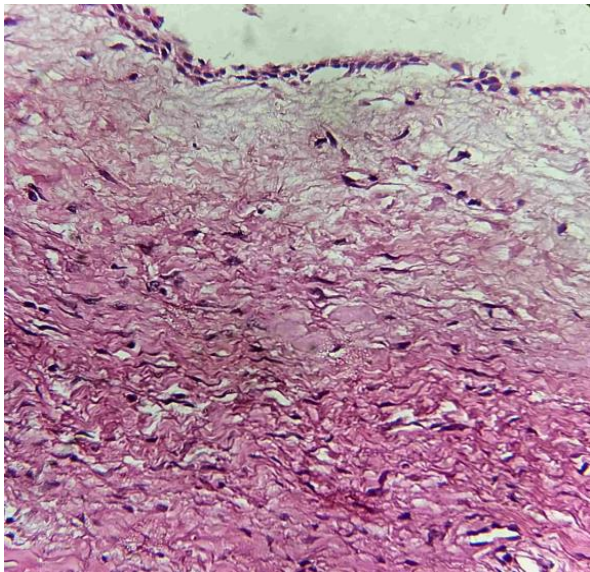


Figure 8: Dentigerous cyst lining (H & E, 10x)

On intraoral examination of the present lesion, the posteromedial portion of the mandibular ramus exhibited mild expansion and tenderness. Orthopantomograph revealed well defined radiolucency of size 5 x 3 cm anteroposteriorly extending from the distal aspect of root remnant of 36 to mandibular ramus

upto the sigmoid notch. Loss of cortical outline of ramus noted anteriorly (Figure 9).

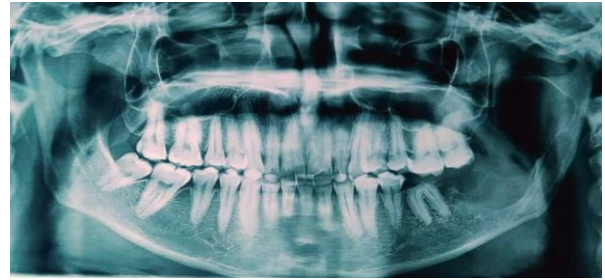


Figure 9: Orthopantomograph revealed well defined radiolucency of size 5 x 3 cm anteroposteriorly extending from the distal aspect of root remnant of 36 to mandibular ramus upto the sigmoid notch

Based on the clinical and radiological features, a provisional diagnosis of Ameloblastic transformation of Dentigerous Cyst was given. Incisional biopsy done from the lesion and specimen was sent for histopathological examination.

The H and E stained sections showed a cyst wall which was lined by non-keratinized epithelium of 2 to 4 cell layers thickness (Figure 10). The epithelium was proliferating into the stroma as tall columnar ameloblast like cells with occasional palisading and stellate reticulum like cells. Within the stroma, there were 2 to 3 ameloblastomatous follicles (Figure 11) and the histopathologic findings were compatible with an ameloblastoma emerging from the dentigerous cyst lining. For subsequent treatment, the patient was referred to the department of oral and maxillofacial surgery.

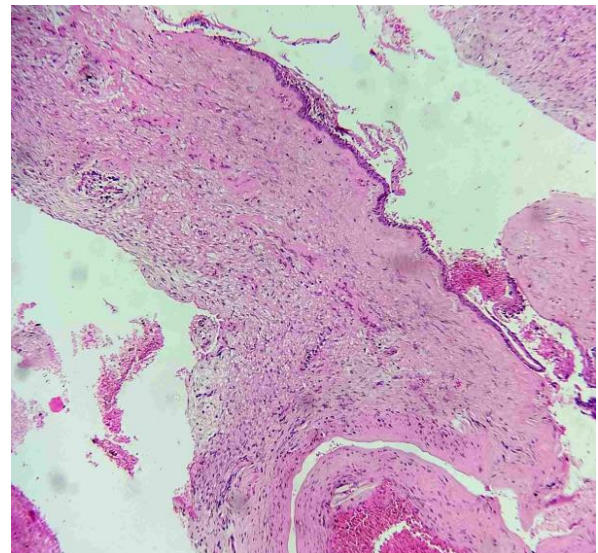


Figure 10: Dentigerous cyst lining (H & E, 4x)

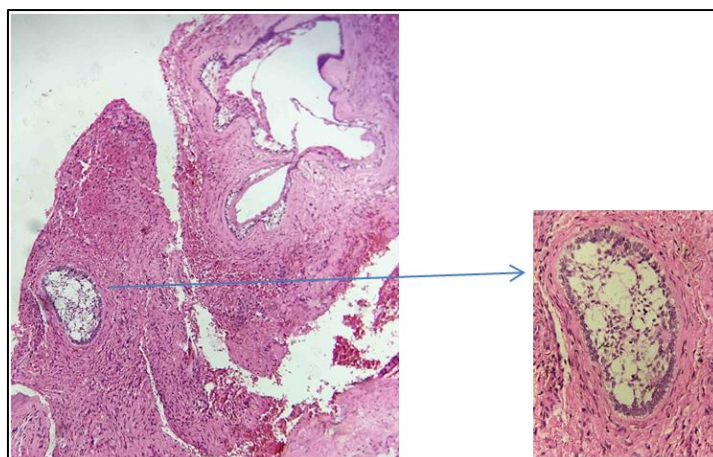


Figure 11: The epithelium is proliferating into the stroma as tall columnar ameloblast like cells with occasional palisading and stellate reticulum like cells and stroma contain ameloblastic follicle (H & E, 4x)

DISCUSSION

Gorlin *et al.*, who reported ameloblastomatous alterations of cysts in nevoid basal cell carcinoma syndrome in 1965, first described the incidence of malignancies emerging from odontogenic cysts [3]. The lining of an odontogenic cyst can transform into odontogenic tumours like ameloblastoma and adenoamatoid odontogenic tumour (AOT), as well as non-odontogenic neoplasms like mucoepidermoid carcinoma and squamous cell carcinoma. However, just a few occurrences of this transformation have been documented in the literature [4].

Dentigerous cyst is often seen between 10 to 30 years of age group and usually associated with unerupted mandibular third molar. Similar findings have been observed in both of the reported cases, which are interestingly associated to the mandibular left third molar. Ameloblastomas are odontogenic epithelial tumours that can develop from the lining of a dentigerous cyst or any other odontogenic epithelial tissue. As a result of this presumption, dentigerous cysts should be managed with caution because they are pre-ameloblastomatous. The next possible reason for believing many ameloblastomas develop from dentigerous cysts is that biopsies of ameloblastomas may be taken of an expanded locule lined apparently by a thin layer of epithelium [1].

A major point of conflict regarding ameloblastoma nowadays is the distinction between ameloblastoma with cystic degeneration and ameloblastoma emerging from a dentigerous cyst. Many authors suggest that the ameloblastic change of the cystic lining is a likely explanation for the pathogenesis of the transformation of ameloblastoma from dentigerous cyst. The presence of an ameloblastomatous epithelial lining of most odontogenic cysts is insufficient to diagnose unicystic ameloblastomas, unless more diagnostic features proposed by Vickers and Gorlin are evident [2]. Based

on various recent studies calretinin is considered as a specific immunohistochemical marker for neoplastic ameloblastic epithelium and serve as a diagnostic tool for differentiating odontogenic cystic lesions from ameloblastic tumours [5].

A study was conducted recently to investigate the expression of p63, CD10 and osteopontin in dentigerous cyst and ameloblastoma to harmonize their expression with neoplastic potentiality. This study concluded that these markers seemed to be up-regulated in dentigerous cysts in the process of transformation to ameloblastoma [6].

The pathogenesis of cyst-tumor transformation till now has considered that the tumors mostly arise from the cyst lining and medical literature does not give attention to the role of odontogenic nests in dentigerous cysts [4]. In the current cases, histopathology was consistent with ameloblastomatous transformation of dentigerous cysts lining. However odontogenic nests are of greater relevance in syndromic cases of keratocystic odontogenic tumor, where they are suspected to be one of the important causes of recurrence. According to Jayanandan M *et al.*, [7] these nests may proliferate and induce changes in the stroma, which is central to the pathogenesis of odontogenic tumors. As a result, even in cases of dentigerous cysts, long-term follow up appears to be recommended.

CONCLUSION

It is critical for the pathologists and clinicians to determine whether a dentigerous cyst is undergoing any ameloblastomatous transformation for proper diagnosis and treatment planning. In future, the use of specific immunohistochemical markers for neoplastic ameloblastic epithelium might be a valuable tool in the diagnosis of odontogenic cystic transformation to tumours.

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