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# Ameloblastomatous transformation in dentigerous cyst: A case report

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ABSTRACT

Mandibular swellings can be caused by a number of benign lesions, which can be of odontogenic or nonodontogenic origin. Lesions include ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumor, central giant cell granuloma, fibro-osseous lesion, and osteoma.[1]

The most common developmental cyst in oral cavity is dentigerous cyst, which accounts for 20% of the developmental cysts of jaws and originates because of accumulation of fluid between reduced enamel epithelium and the tooth crown. Hence, it usually is associated with impacted or unerupted tooth.[2] Ameloblastoma is a locally aggressive odontogenic neoplasm accounting for approximately 10% of all the neoplasms arising in jaws.[3] Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts. The goal of treatment for ameloblastoma is to achieve complete excision and appropriate reconstruction.

We present a case of a large dentigerous cyst transforming into an ameloblastoma.

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Key message:

> Transformation of cystic lining of dentigerous cyst into ameloblastoma is a rare occurrence. The surgeon and the oral pathologist should endeavor for a thorough curettage and serial sections of the lining, respectively, to comprehensively treat the patient.

## 1. Case history

A 15-year-old male patient was referred to us in June 2012 with complaints of swelling over right side of face since 2 months [Fig. 1].

On examination, a bony hard swelling with crepitation at places was found on the right side of face extending from body of mandible (Rt) to sigmoid notch (Rt) involving the ramus extensively. Intraoral examination showed 48 was missing. On palpation, buccal cortex was found expanded with perforation at places. Lingual cortex was normal except at 46–47 region where marked expansion was noticed. No intraoral discharge of fluid was observed. The cystic fluid was aspirated from the swelling, which was sent for fine-needle aspiration cytology (FNAC) and protein estimation. The protein levels were found to be within normal range.

The orthopantomogram (OPG) X-ray [Fig. 2] image provided an overview of an extensive progressive cystic lesion extending from sigmoid notch of right mandible to body of the mandible and involvement of buccal cortex alone at most

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**Fig. 1 – Swelling on right side face.**

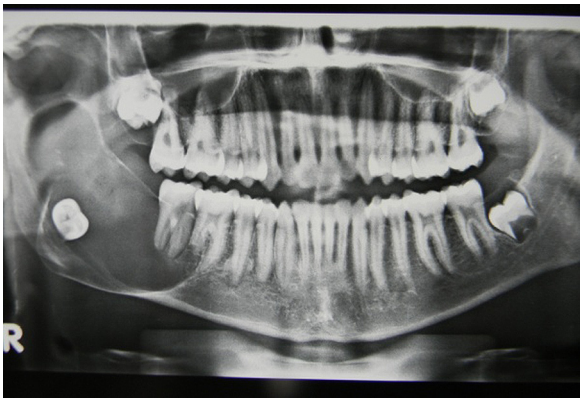


**Fig. 2 – OPG showing the radiolucency.**

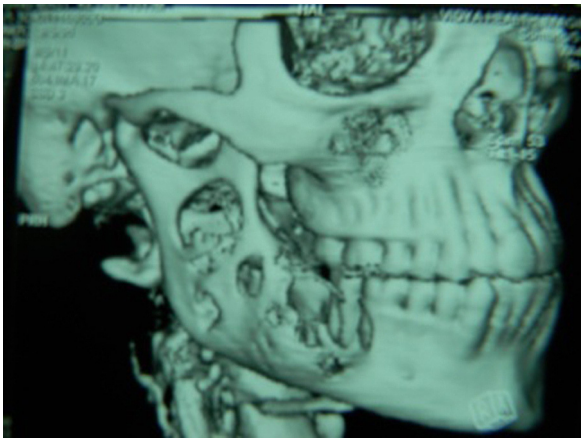


**Fig. 3 – CT scan.**

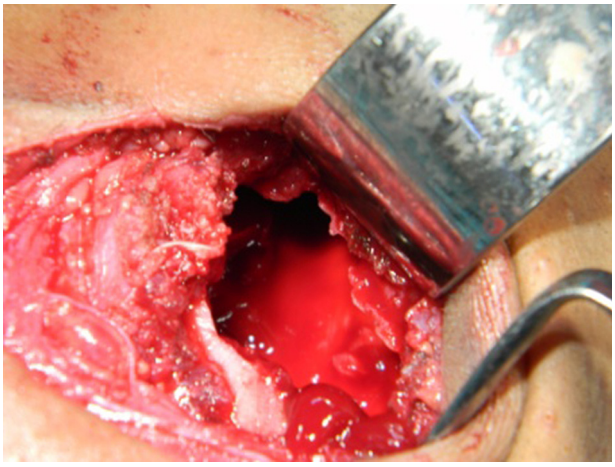


**Fig. 4 – Risdon approach.**

places. However, ramus (Rt) showed multiple perforations. Resorption of distal root of 46 was also noticed.

On review of the CT [Fig. 3] along with clinical findings, we found the involvement of lingual cortex of body of mandible along with perforation at multiple places of ascending ramus and body of the mandible.

## 2. Maxillofacial surgical procedure

Incisional biopsy on the affected region was performed and histopathologic report confirmed dentigerous cyst mandible (Rt).

The patient was taken up for complete enucleation of cystic lesion, and peripheral ostectomy was performed under general anesthesia. The cyst was approached extraorally using Risdon approach [Fig. 4]. The thin flakes of buccal cortical bone were removed, and the cystic lining along with impacted 48 was removed in toto. A small portion of healthy bone (4 mm) was also removed at the mesial end of the lesion after extraction of 46 and 47. All soft tissues in contact or overlying the lesion were excised. However, the compact bone at lower border of mandible and ascending ramus along with periosteum were conserved. Carnoy's solution was applied at the wound with peanut swab. At the mesial end of the lesion, electrocautery was applied at the tooth root socket and adjacent area. A suction drain was placed before wound closure. The sample was sent for histopathologic evaluation with its border marked [Fig. 5].

Postoperative recovery was uneventful with administration of intravenous antibiotic. The suction drain was removed after 48 h, and the patient was discharged 7 days after the suture removal.

The biopsy result showed a cystic lesion with thin non-keratinized stratified squamous epithelium with no rete processes, and the subjacent connective tissue was devoid of any inflammatory cells, consistent with the diagnosis of which corresponded to the cyst lining the ramus and body of the mandible [Fig. 6] whereas at mesial border, cystic lining proliferation into the lumen was noticed [Fig. 7]. Ameloblastic islands showing reversal of polarity of nuclei and cytoplasmic vacuolations were also observed [Fig. 8]. Histopathologic findings were consistent with those of an ameloblastoma arising within a dentigerous cyst lining and associated with an unerupted tooth.



Fig. 5 – Specimen.

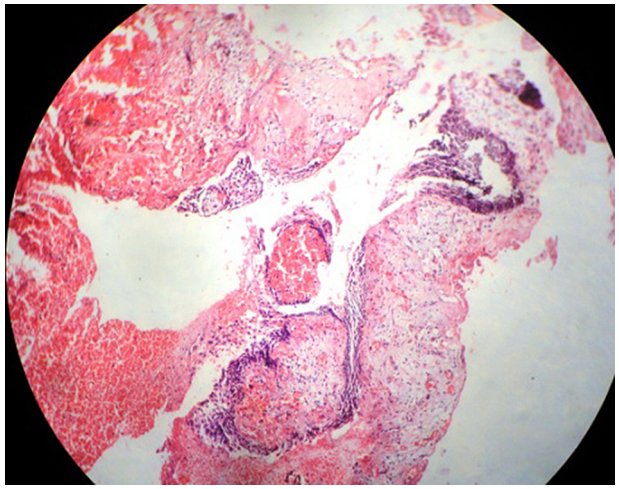


Fig. 7 – Cystic proliferation in the lumen.

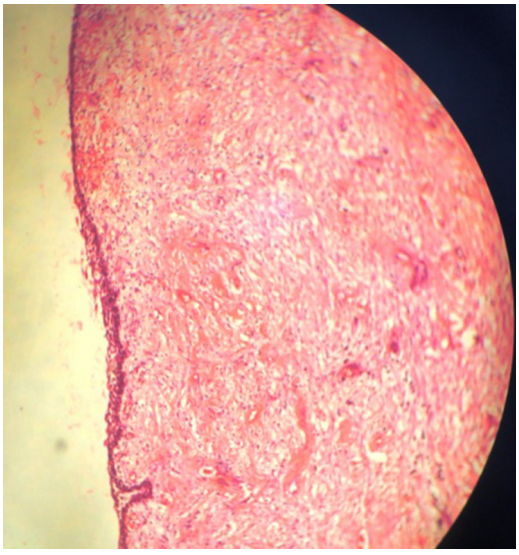


Fig. 6 – Features of dentigerous cyst.

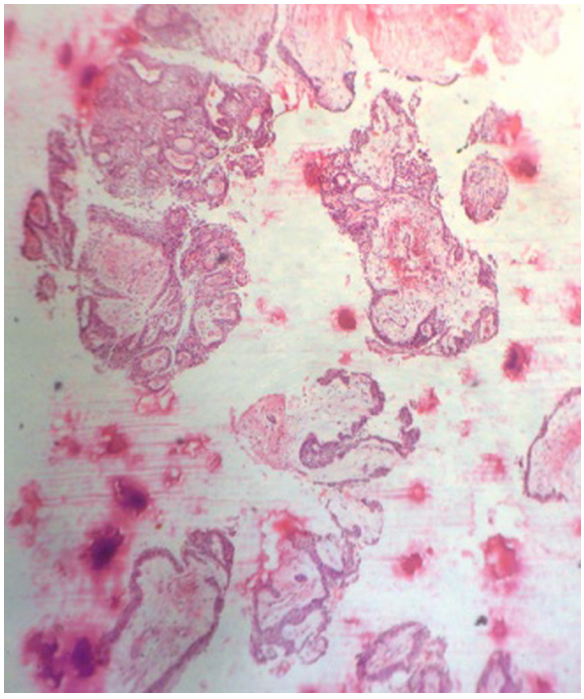


Fig. 8 – Ameloblastic islands.

The patient is being routinely followed up. As on June 2014 (24 months after the operation), no signs of recurrence have been seen, and the patient is taking active part in his routine life [Fig. 9].

## 3. Discussion

Though dentigerous cysts are more common among 30- to 40-year-olds,[4] its occurrence in children, as in our case, is not uncommon. Occurrence of ameloblastoma is predominant among 20- to 40-year-olds with almost equal propensity of occurrence in both genders.[4] Ameloblastomas are locally aggressive and rarely metastasize.[5]

Impacted and unerupted molars are associated with various odontogenic cysts and tumors. Whereas dentigerous cysts are the most common cysts among them (93%), the occurrence of ameloblastomas is not so frequent (0.41%).[4]

Radiographic examination is an important tool in the investigation of a missing tooth. An intraoral periapical (IOPA) or occlusal radiograph provides important information. OPG provides detailed overview of the extent of lesion, root resorption, relation of teeth adjacent to cysts, and so on whereas CT is one of the most useful tools for differential diagnosis of the lesion.[6]

Recently, a study was conducted to investigate the expression of CD10 and osteopontin in dentigerous cyst and ameloblastoma to correlate their expression with neoplastic



**Fig. 9 – Post-op recovery.**

potentiality. It was concluded that high CD10 and osteopontin expression may indicate the neoplastic potentiality of certain areas.[7]

Various contradictory theories about the development of unicystic ameloblastomas (UAs) have been proposed. Although some authors suggest that UAs develop by cystic degeneration of solid ameloblastomas, there are certain indications that they may develop by mural and/or luminal ameloblastomatous change in a preexisting cyst.[8] It has also been shown that in UAs, a coexistence of nonneoplastic and neoplastic epithelium is possible.

A histologic distinction between UAs and certain nonneoplastic odontogenic cysts can be difficult.[9] It has been suggested that six radiographic patterns for UAs can be identified ranging from well-defined unilocular to multilocular.

Many attempts have been made to establish specific immunohistochemical markers for ameloblastomas.[10] For example, it has been suggested that differences in the expression of cell surface carbohydrates with blood group specificity may be helpful to distinguish ameloblastomas from odontogenic cysts,[10] although this hypothesis has not been confirmed by other authors. Another study evaluated the proliferating cell nuclear antigen (PCNA) in the cystic tumor lining of UA and found significantly higher number of PCNA-positive cells in UA than in dentigerous cyst linings. Recently, it has been suggested that calretinin is a specific immunohistochemical marker for neoplastic ameloblastic epithelium and may serve as a diagnostic tool for differentiating cystic odontogenic lesions from ameloblastic tumors.[9] However, till now, no general recommendations exist.

Odontogenic cell nests are frequently found in the connective tissue associated with odontogenic cysts. Some of them may show palisading of basaloid cells located at the outer circumference while other signs of true ameloblastic differentiation, such as basal cell hyperchromatism, polarization of the basal cell nuclei, or vacuolation of the basal cell cytoplasm, are lacking.

In our case, extensive step sectioning showed rather large odontogenic cell nests intraluminally on the mesial side of the lesion. As these cell nests were intraluminal, it could be assumed that they were completely removed along with the enucleation of the cyst.

It has been suggested that histologic examination is the sine qua non for differentiating between odontogenic cysts and UAs. However, both clinical and radiological findings contribute to the diagnosis. In future, the use of specific immunohistochemical markers for UAs might be a valuable tool in the differential diagnosis of dentigerous cysts and ameloblastomas.

This case was surgically treated by following a routine protocol of the management of dentigerous cyst by complete enucleation.[6] The area where the cyst had undergone ameloblastic changes was dealt conservatively by preserving maximum soft and hard tissues after keeping safe margin. Though one-step procedure of immediate reconstruction of jaw in ameloblastoma cases has been contemplated, it was decided not to carry out immediate reconstruction keeping in mind the academic and social obligation of the patient.

## Conflicts of interest

The authors have none to declare.

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