Case Report

Ameloblastoma arising in the wall of dentigerous cyst: Report of a rare entity

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Abstract

Dentigerous cyst (DC) is a developmental odontogenic cyst that encloses the crown of an unerupted tooth by expansion of its follicle with accumulation of fluid between the reduced enamel epithelium and the tooth crown and is attached to the neck of the tooth. The lining of DCs shows a potential for neoplastic transformation to ameloblastoma, squamous cell carcinoma, and mucoepidermoid carcinoma. Here, we report a rare case of an ameloblastoma arising in the wall of a DC.

Keywords: Ameloblastoma, dentigerous cyst, neoplastic transformation

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INTRODUCTION

The odontogenic cysts with the neoplastic potential include dentigerous cyst (DC), odontogenic keratocysts, calcifying odontogenic cysts, glandular odontogenic cysts, and radicular cysts. Among the odontogenic cysts, neoplastic transformation is highest in odontogenic keratocyst and DC.[1] DC is the most common type of odontogenic cyst which is formed by the accumulation of fluid between the reduced enamel epithelium and the tooth crown and clinically associated with unerupted tooth most commonly unerupted mandibular third molar, maxillary canine, and mandibular premolars. Radiographically, unilocular radiolucency with well-defined sclerotic margins surrounding crown of an unerupted tooth is noticed.[2] Till date, very few cases of ameloblastoma arising in the wall of DC have been reported similar to the present case which was reported in 19-year-old male patient.

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CASE REPORT

A 19-year-old male reported to outpatient department complaining of an asymptomatic swelling on the left side of face for 1 year. The swelling was initially peanut in size which gradually increased to the present size. Past dental history revealed extraction of decayed 36. General physical examination revealed no abnormality.

Extraoral examination revealed facial asymmetry with a diffuse swelling on the left side of face, extending from ala-tragus line to lower border of mandible measuring approximately of size 5 cm × 6 cm [Figure 1]. On palpation, all the inspection findings were confirmed, and the swelling was tender and firm to hard in consistency. Single left submandibular lymph node is palpable which is firm, tender measuring approximately 0.5 cm × 0.5 cm.

Intraoral examination revealed a swelling with smooth surface, obliterating buccal vestibule, and extending

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anteroposteriorly from 35 to alveolar ridge of 37, followed by ascending ramus till occlusal level of 26 and 27. Buccal and lingual cortical expansion was also noticed. On palpation, the swelling is soft at 35 and retromolar region whereas the rest of the lesion is firm in consistency. Missing teeth are present in relation to 28, 36, 38, and 48 and Grade I mobility in relation to 37.

Orthopantomograph revealed multilocular radiolucency on left side extending from 35 till condylar process. Perforation of lingual cortex at 37 region and root resorption in relation to 37 with impacted 38 was also noticed [Figure 2]. Computed tomography scan revealed buccal and lingual cortical expansion with cortical perforation at 37 region [Figure 3]. Based on the clinical and radiological features, a provisional diagnosis of DC was given. An incisional biopsy was sent for histopathological examination, and the findings revealed the features of DC. The complete lesion was surgically removed along with normal tissue margins surrounding the lesion [Figure 4], and the excised specimen was sent for histopathological examination.

Macroscopically, a hemimandibulectomy specimen with 35 and 37 teeth which is grayish white in color measuring approximately 12 cm × 7 cm was noticed. The specimen was roughly rectangular in shape, hard in consistency with smooth surface, and well-defined borders [Figure 5a]. The specimen was cut into two halves; in one half of the bony specimen, an impacted third molar was noticed. Inside, the bony cavity gelatinous material was present which is light green in color [Figure 5b].

Histopathology of excisional biopsy revealed cystic cavity lined by cystic epithelium resembling reduced enamel epithelium. The epithelial lining revealed tall columnar basal layer, reversal polarity of hyperchromatic nucleus, degenerating stellate reticulum-like cells, and focal areas of subnuclear vacuolization. The underlying connective tissue stroma shows odontogenic epithelial rests with focal areas

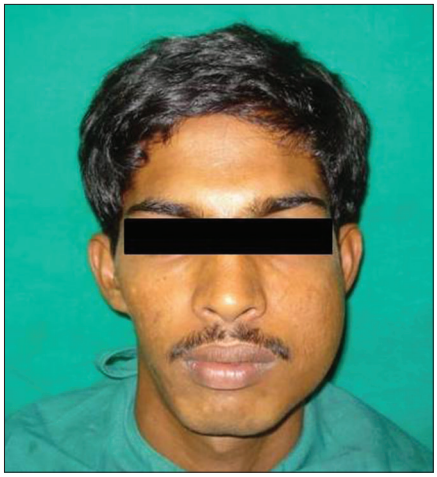

Figure 1: Extraoral facial asymmetry on left side of face

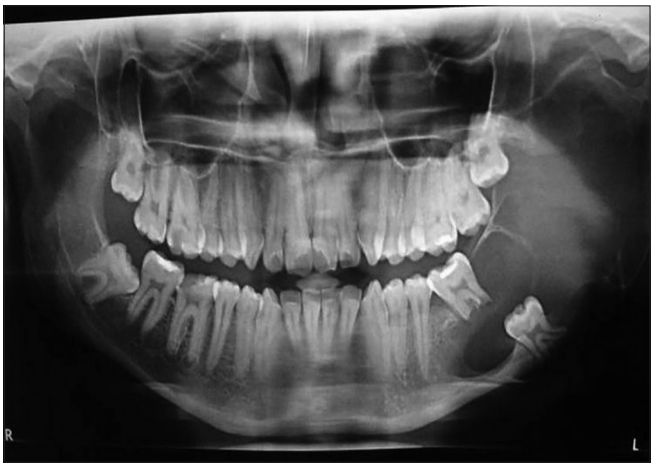

Figure 2: Orthopantomograph revealed multilocular radiolucency on left side extending from 35 to condylar process

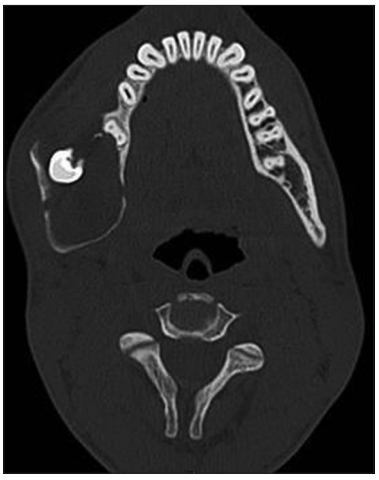

Figure 3: Computed tomography scan revealing buccal and lingual cortical expansion with cortical perforation at 37 region

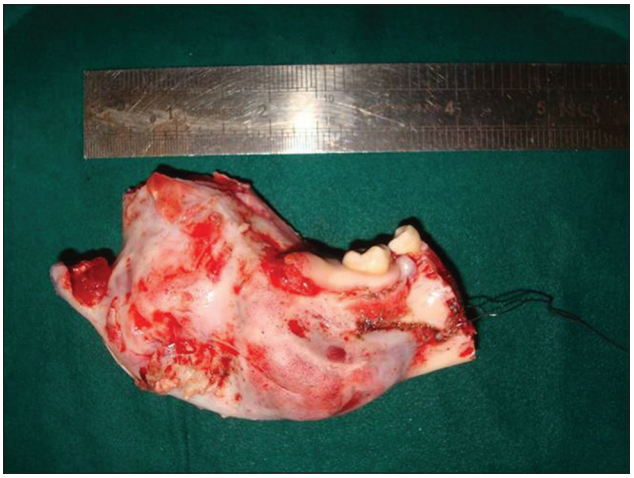

Figure 4: Surgically excised specimen

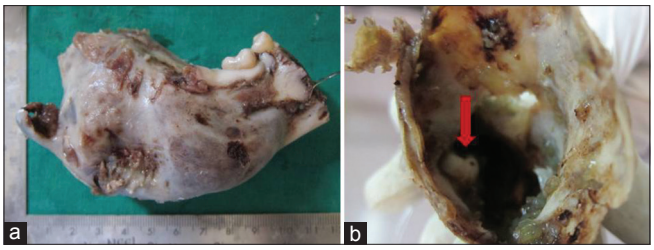

Figure 5: (a) Macroscopic specimen, (b) Specimen with impacted tooth

of follicular ameloblastomatous islands [Figure 6]. Based on these findings, a final diagnosis of DC transforming into ameloblastoma was given.

DISCUSSION

The DC is the second most common odontogenic cyst, after radicular cyst and accounts for >24% of jaw cysts.[3] It presents mostly in the two to third decade of life. DCs are mostly asymptomatic and detected on routine radiographic examination. The outgrowth of the cyst also causes resorption of adjacent tooth. Histological examination reveals a 2–4 cell thickness, flat or cuboidal epithelium usually nonkeratinized, with a thin fibrous cyst wall. DCs are diagnosed based on a combination of radiographic and histopathological features.[4]

DCs are believed to be formed from the fluid accumulated between reduced enamel epithelium and tooth crown and results in the expanded follicle beyond the 3 mm normal diameter because of which they are usually associated with impacted or unerupted teeth. Few cases reported in literature show the neoplastic potential of the epithelium of DCs to ameloblastoma, epidermoid carcinoma, and mucoepidermoid carcinoma.[5,6]

An ameloblastoma is a locally aggressive benign epithelial odontogenic tumor, commonly arising from the mandible mostly in third to fifth decades. It occurs commonly in molar-ramus region of mandible. Ameloblastoma may arise from the remnants of dental lamina and enamel organ or from the basal layer of oral epithelium as well as the epithelium of DC. Radiographically, it appears as uni/multilocular radiolucency with a typical honeycomb or soap bubble appearance.[7,8]

According to the World Health Organization, ameloblastomas can be classified into four groups: solid/multicystic, extraosseous/peripheral, desmoplastic, and unicystic.[9] Histopathologically, it occurs in six patterns: follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic type.[10] Vicker and Gorlin suggested that the following features as representative

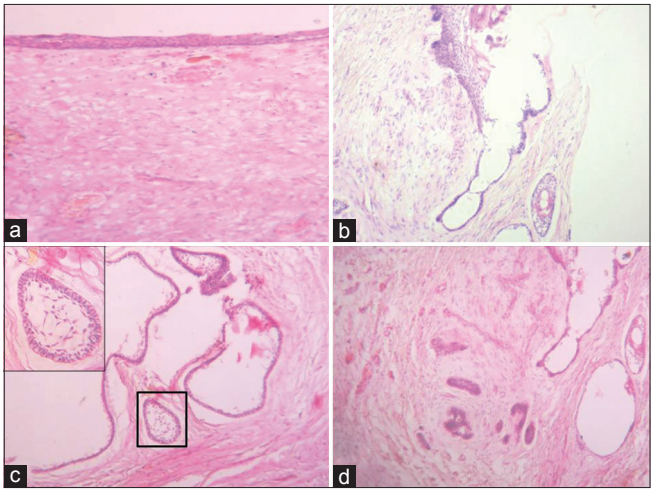

Figure 6: (a) Dentigerous cyst epithelial lining, (b) Lining revealing V and G criteria, (c) Ameloblasotmatous follicle within the stroma, (d) Ameloblastomatous islands in the stroma along with cyst lining

of early ameloblastomatous changes include an epithelial lining of which parts may show transformation to cuboidal or columnar basal cells with hyperchromatic nuclei, nuclear palisading with polarization, cytoplasmic vacuolization with intercellular spacing, and subepithelial hyalinization.[11]

It has been reported that the epithelium of odontogenic cyst may be transformed into benign odontogenic tumors such as ameloblastoma, adenomatoid odontogenic tumor, and nonodontogenic malignant tumors such as epidermoid and mucoepidermoid carcinoma. The frequency of such neoplastic transformation is very low.[12,13]

According to shear, the most probable reasons are an ameloblastoma has similar clinical and radiographic features to DC and microscopic features conclude that the ameloblastoma arising from DC. Next, reason reveals that a biopsy taken from an expanded locule may be an ameloblastoma developing in the epithelial lining of DC. Final reason states that isolated islets or follicles of ameloblastomatous epithelium are found in the cyst wall at some distance from the epithelial lining.[14] In our case, the occurrence of DC cystic lining away from neoplastic epithelium and the unerupted tooth concludes that DC is turning into ameloblastoma.

Various proliferative studies and immunohistochemical demonstration of proliferative markers suggest that increased cell proliferation plays a role in the development of odontogenic cyst (e.g., DC) and neoplastic tumors (e.g., ameloblastoma). This increased cell proliferation can result from disruption in cell cycle, mutations in oncogenes, or tumor suppressor genes.[15,16]

CONCLUSION

The origin of ameloblastoma from DC is still controversial. Our present case of an ameloblastoma arising from a DC is a rare entity that unfolds the histogenesis of ameloblastoma. This case highlights the neoplastic potential of DCs and importance of careful histopathological examination of the whole specimen with multiple sectioning.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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