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CASE REPORT PAPER

Aggressive plexiform mandibular ameloblastoma arising from a dentigerous cyst: A case report

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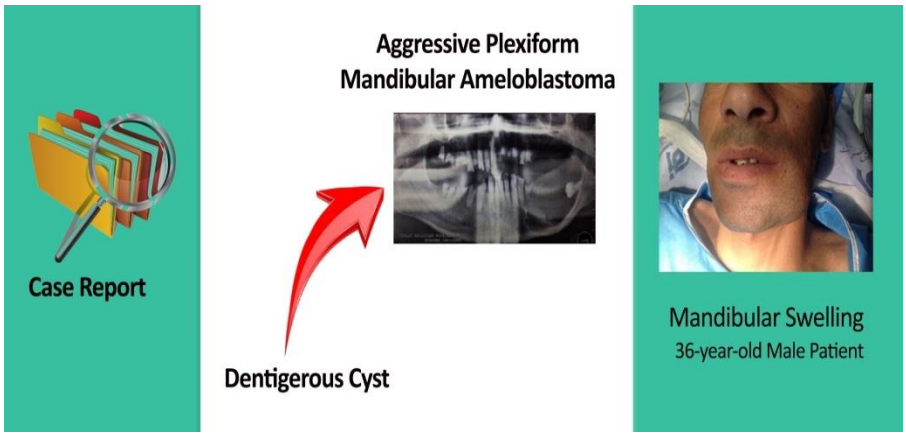
Highlights

- Dentigerous cyst is the most common type of developmental odontogenic cysts.
- Ameloblastoma is the most common clinically significant odontogenic tumor.
- Ameloblastomas are tumors of odontogenic epithelial origin.
- Ameloblastomas can arise from the epithelial lining of odontogenic cyst.

Graphical Abstract



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Abstract

Among odontogenic cysts, dental cysts (DC) and among odontogenic tumors, ameloblastoma are the most common. Odontogenic tumors such as ameloblastoma can develop from the cyst lining of the dentigernary cyst and also the odontogenic keratocyst. DC is the most common form of odontogenic cyst, which results from the accumulation of fluid between the reduced enamel epithelium and the tooth crown and is clinically, associated with an unerupted tooth, usually an unerupted mandibular third molar, maxillary canine, and mandibular premolar. Radiographically, unilocular x-ray fluorescence is noted with well-defined sclerotic margins surrounding the crown of an unerupted tooth. Ameloblastoma is a neoplasm classified as a benign epithelial odontogenic tumor of the jaw. 70% of ameloblastomas develop in the molar-ramus region of the mandible and are occasionally associated with an unerupted third molar. Histologically, most ameloblastomas have a follicular or plexiform pattern, although basaloid, granular, or desmoplastic cellular changes may also occur. This study presents a case of an aggressive plexiform mandibular ameloblastoma arising from the cyst lining of a dentigerous cyst in a 36-year-old male patient.

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Introduction

DC is the most common type of odontogenic cyst, and it is formed by the accumulation of fluid between the reduced enamel epithelium and the tooth crown. It is most commonly found in people who haven't had their teeth come in, like the mandibular third molar, the maxillary canine, and the mandibular premolars. The crown of an unerupted tooth will have a unilocular radiolucency and well-defined sclerotic edges, as seen on radiographs. The cyst is walled by two to four layers of odontogenic epithelial cells with a flat connective tissue interface, and some goblet cells (cells with foamy cytoplasm) may be seen among them histopathologically dentigerous cyst. Among the benign epithelial-odontogenic tumours of the jaws, ameloblastoma is one of the most common types of neoplasia (1). The molar-ramus area of the jaw is the site of 70% of ameloblastomas, and an unerupted third molar tooth may be involved in certain cases (2). Histologically, the majority of ameloblastomas exhibit a follicular or plexiform appearance; however basaloid, granular, or desmoplastic cellular alterations may also be seen (3).

Case Presentation

A 33-year-old male patient presented to the clinical infirmary of Mashhad dentistry school in Jan 2018, complaining of edoema on the left side of the jaw. In view of the disease's history, the discomfort began two years ago with a minor swelling, and the teeth on the same side were taken without radiography due to the cause of pain. However, each time, the pain persisted and only a slight reduction in swelling occurred as a result of the antibiotics taken, but the swelling returned, and when the swelling was significantly increased, panoramic radiographs were taken and radiolucency was noted in the left area of the mandible (Figure 1).

On the radiograph, can see a mixed pericoronal lesion with a well-defined cortical boundary which extending from the distal of tooth number 34 extending up to the ascending ramus, with the pericoronal lesion extending into the impacted tooth 38. Severe expansion is seen in the vertical dimension, which also causes mandibular canal displacement, but perforation of the lower cortex is not seen although the buccal cortex have been eroded in this view; and a pseudo-septum is also seen in the lower part of the lesion, which is probably due to the lesion expansion (Figure 2).

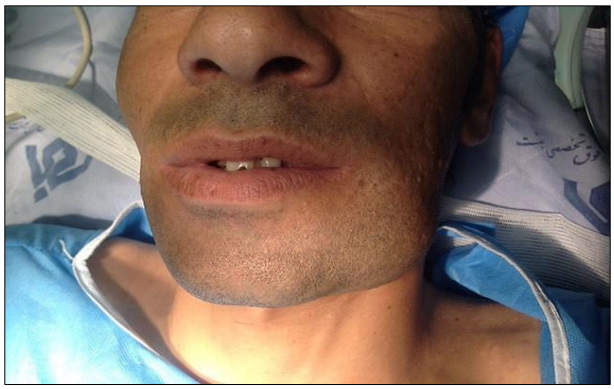

Figure 1. Patient profile with mandibular swelling on the left side.

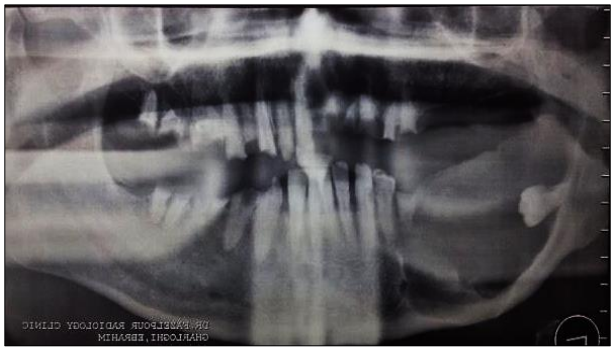

Figure 2. Panoramic X-Ray showing an expansive radiolucency on the left side of mandible area of teeth #34 up to ascending ramus.

A series of small calcifications are seen on the CT, which may be trabeculae of bone. In buccolingual dimension, severe expansion, perforation of the buccal cortex, and removal of the lingual cortex can be observed. Considering these characteristics, it seems that the differential diagnosis is ameloblastic fibro-odontoma, COC, CEOT, and the best diagnosis is unicystic ameloblastoma (Figure 3).

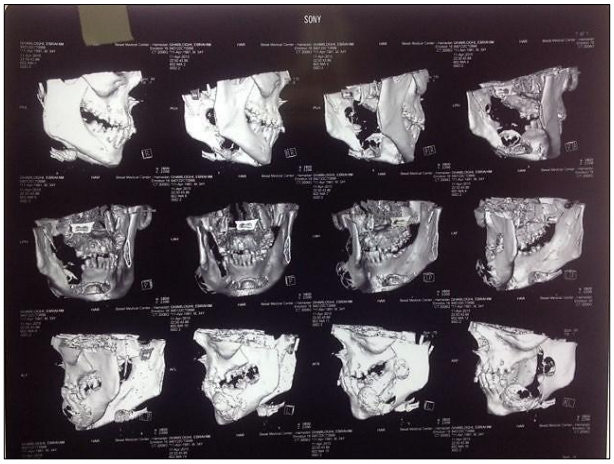

Figure 3. CT scans showing expansive lesion on the left side of mandible.

In order to be able to prepare a good histopathologic slide, first the hard tissue is separated from the soft tissue and the hard tissue is prepared separately. Hemi mandibulectomy consisting of mandibular ascending ramus and condyle and complete coronoid (Figures 4 and 5).

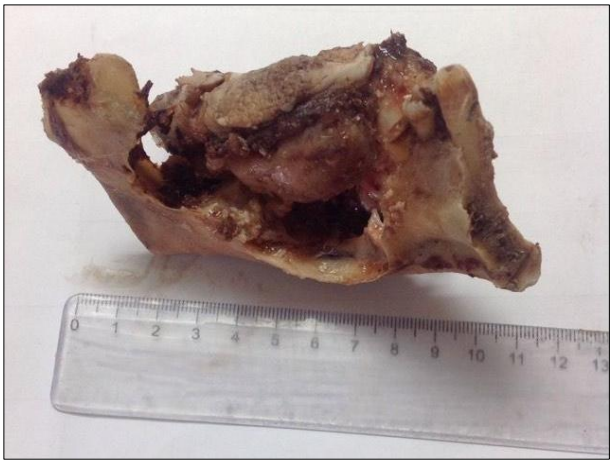

Figure 4. Gross specimen.

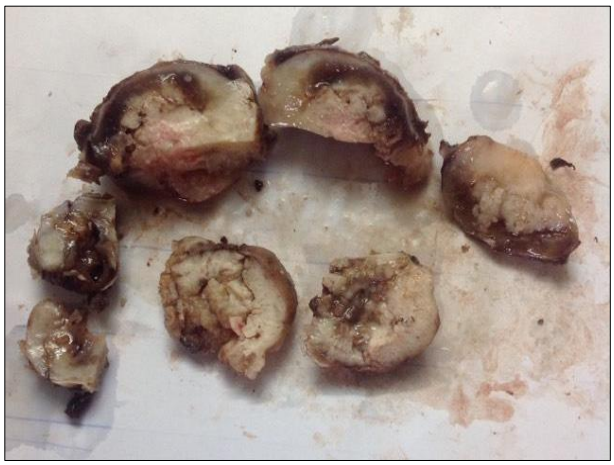

Figure 5. Cut surface of gross specimen.

Microscopic evaluation shows odontogenic epithelial tumor consisting of strands, codes and networks of columnar (ameloblast-like) cells with reverse polarity and some degree of pleomorphic changes that are surrounding stellate shape cells which are loosely arranged and are located behind these ameloblast like cells which is extended to the soft of the alveolar mucosa (Figure 6). There is also a cyst consists of two to four layers of odontogenic epithelial cells with flat connective tissue interface and few goblet cells (cells with foamy cytoplasm) among them, in the vicinity of this odontogenic tumor (Figures 7 and 8). Diagnosis of plexiform ameloblastoma arising from cyst lining of dentigerous cyst have been considered for this microscopic finding.

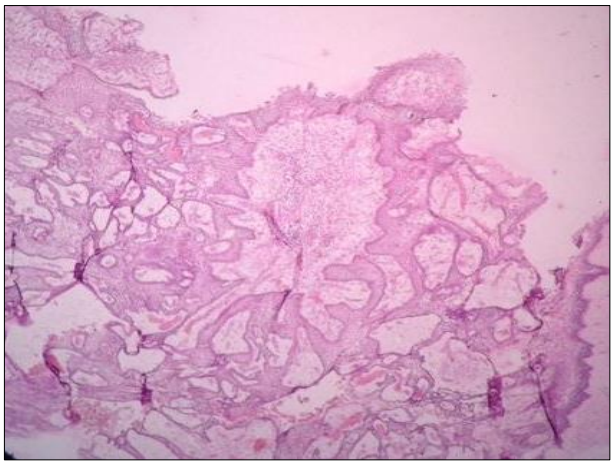

Figure 6. Microscopic picture showing networks of plexiform ameloblastoma (X40).

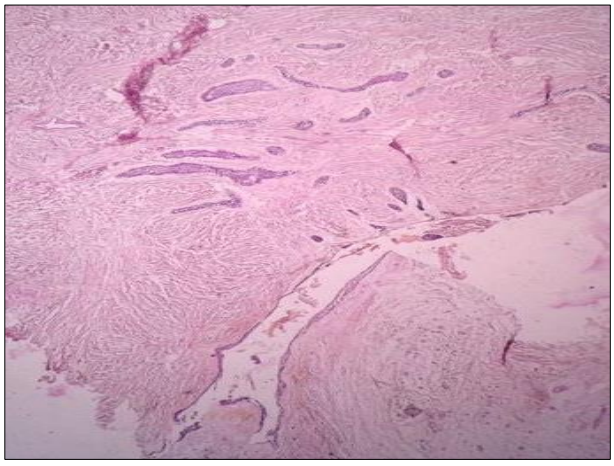

Figure 7. Low power photomicrograph showing odontogenic (dentigerous) cyst and islands of ameloblastic transformation (X40).

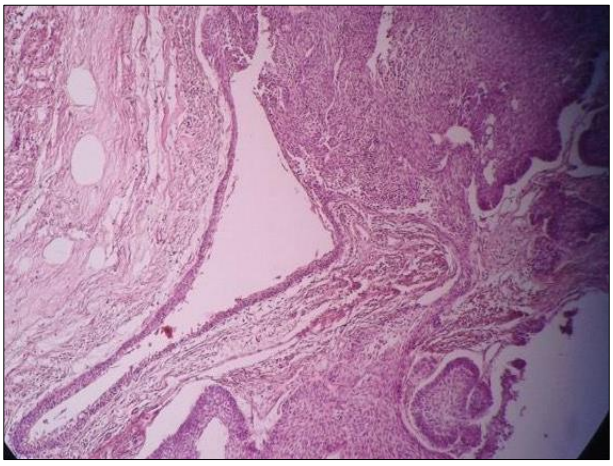

Figure 8. Medium power photomicrograph showing odontogenic (dentigerous) cyst and islands of ameloblastic transformation (X100).

The patient was advised to attend regular follow-up visits every six months. One year after surgery, there were no signs of recurrence.

Discussion

Dentigerous cysts (DC), odontogenic keratocysts, calcifying odontogenic cysts, glandular odontogenic cysts, and radicular cysts are all examples of odontogenic cysts having neoplastic potential. DC (dentigerous cysts) and odontogenic keratocysts had the greatest risk of neoplastic transformation among odontogenic cysts (4). Dentigerous cysts are the most often seen source of pericoronal radiolucency in impacted teeth. They are often asymptomatic and are discovered through regular dental radiographs (5). Until now, relatively few instances of ameloblastoma have been identified that developed from the wall of dentigerous cysts identical to the one described here. Ameloblastoma is the most clinically significant odontogenic epithelial malignancy. It may arise from remains of the dental lamina of the growing enamel organ, which forms the tooth epithelial lining of an odontogenic cyst, or from the oral mucosa's basal cells. They are often slow-growing, locally damaging, and have a benign development history (6). They are monocystic or multicystic radiolucencies with a honeycomb or soap bubble look on radiography (7). It is uncommon in children under the age of ten, accounting for around 10-15% of all recorded cases (8). These lesions often exhibit themselves clinically and radiographically in three distinct ways: a) Common solid or multicystic (about 75-86% of all cases); b) Unicystic (approximately 13-21% of all instances); c) Peripheral or extraosseous (about 1-4% of all cases) (9). Intraosseous multicystic ameloblastoma may occur at any age. Between the third and seventh decades of life, tumours are approximately equally prevalent. There is no preference for one gender over another.

According to certain research, black individuals have a greater prevalence. Around 80-85% of traditional ameloblastomas are located in the mandible, most often in the ascending molar and ramus areas. A typical clinical symptom is a painless swelling or enlargement of the jaw. If left untreated, the lesion may develop slowly but steadily until it reaches a significant size. Even in big tumours, pain and paresthesia are infrequent (6). Multicystic ameloblastoma, whether solid or intraosseous, has a strong proclivity for cystic alterations. Cysts may be seen only under a microscope, or they may be multiple enormous cysts that comprise the majority of the tumour. The most prevalent patterns are follicular and plexiform. Acantomatous, granocellular, desmoplastic, and basal cell cysts are less frequent histological patterns (10). According to several researches, uni-cystic ameloblastomas constitute for 10 to 15% of all intraosseous ameloblastomas (11). The majority of unicystic ameloblastomas occur in the mandible, most often in the posterior portions. Often, the lesion is asymptomatic, however big lesions may induce painless jaw swelling. The lesion often manifests as a radiolucency with distinct borders that surrounds the unerupted third molar in the jaw and clinically mimics a dentigerous cyst in many cases (6). Peripheral ameloblastoma is rare, accounting for around 1 to 4% of all ameloblastomas (12, 13). These lesions resemble intraosseous forms histopathologically. Peripheral ameloblastomas often present clinically as a painless, non-ulcerative, sessile or pedunculated lesion in the gingiva or alveolar mucosa. Curettage, enucleation with curettage, or aggressive surgery is used to treat meloblastomas (14, 15). In this instance, radical mandibular resection (Surgery to remove a tumor and a large amount of normal tissue surrounding it) was contemplated, and three, six, and twelve-month follow-ups revealed no additional tumour development or involvement.

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Conclusion

Since Ameloblastoma can arise from wall of dentigerous cyst a complete cyst removal together with impacted teeth should be considered and in case of Ameloblastoma as they are locally aggressive lesions and

can recure after marginal resection or conservative surgery a total block resection should be considered for treatment.

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