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RESEARCH ARTICLE

DENTIGEROUS CYST VERSUS UNICYSTIC AMELOBLASTOMA – A DIAGNOSTICALLY DIFFICULT SITUATION

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ABSTRACT

Ameloblastoma is the 2nd most common odontogenic tumor following odontoma. The tumor is often asymptomatic and commonly seen in mandible but it may also develop in maxilla. Unicystic ameloblastoma clinically and radiographically presents a similar and confusing picture as of Dentigerous cyst when it is associated with tooth. Histopathological examination is essential to diagnose such cases. We report a case of young male with a radiolucent lesion in left maxilla associated with impacted 3rd molar. Surgical removal of lesion was performed, with differential diagnosis of dentigerous cyst. However, histopathological examination reveled ameloblastoma over dentigerous cyst.

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INTRODUCTION

Ameloblastoma only accounts for only 1% of all oral tumours but in day today clinical practice it is the most common type of odontogenic tumour which we come across (Li and Ktano, 1997). Ameloblastoma believe to be originated from various sources such as residual epithelium of tooth forming apparatus, such as epithelial cell rests of malassez, epithelium of odontogenic cyst, epithelium of enamel organ, basal cells of surface epithelium and heterotopic epithelium from extraoral sites such as pituitary gland (Singh *et al*., 2011). Based on the world health organization (WHO) classification of ameloblastoma, there are four types of ameloblastoma: multicystic, peripheral, desmoplastic and unicystic ameloblastoma (Kramer *et al*., 1992). In this report, a case of unicystic/mural ameloblastoma arising from dentigerous cyst in maxilla of a 15 year old male patient.

CASE REPORT

A 15 year old male patient reported to outpatient department with chief complaint of swelling over left side of face since two years which was gradually increasing in size. Medical history reveals no systemic diseases. Extraoral examination revealed facial asymmetry on the left side of face (Fig.1). The skin overlying swelling was normal. The extraoral swelling was ill-defined, painless and approximately 4x4 cm in size. The swelling was hard in consistency and no fluctuation was present. Intraoral examination revealed swelling in left maxillary quadrant vestibular area extending from left canine to second molar area causing complete obliteration left maxillary vestibule. Egg shell crackling was present bucally but not palatally on palpation. Orthopantamogram revealed a unilocular radiolucent lesion extending from maxillary left second premolar to maxillary left second molar along with impacted third molar attached inferiorly to roots of teeth and also show resorption root of first and second molar and lesion extending superiorly in the maxillary sinus (Fig.2). CT finding revealed monocortical buccal expansion and also expansion of anterior wall of maxillary sinus. CT also revealed haziness and complete obliteration of left maxillary sinus and it also contained an impacted tooth (Fig. 3). Based on clinical and radiological feature a pre-operative diagnosis of dentigerous cyst associated with impacted third molar was established and planned for exisional biopsy under local anaesthesia. The patient was operated under local anaesthesia using intraoral approach and complete enucleation of cystic lesion was done along with removal of impacted third molar (Fig. 4). After removal of lesion (Fig. 5), gauze soaked with betadine is packed into hollow cavity and primary closure done on the second post-operative day after removal of gauze. The cystic lesion was sent for histopathology. There after patient was recalled on follow-up on regular basis. The histopathological

examination revealed ameloblastoma over dentigerous cyst (Fig. 6).

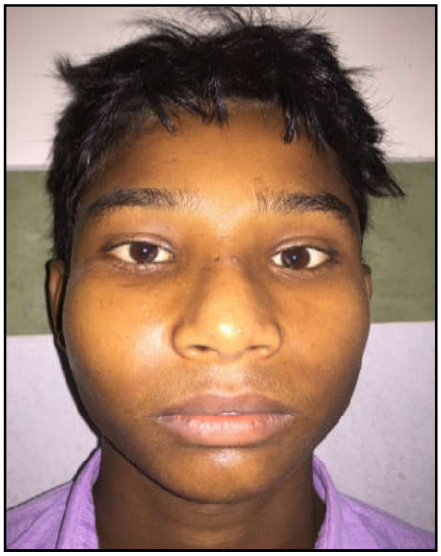


Fig.1. Extra-oral photograph showing facial asymmetry and swelling over left side

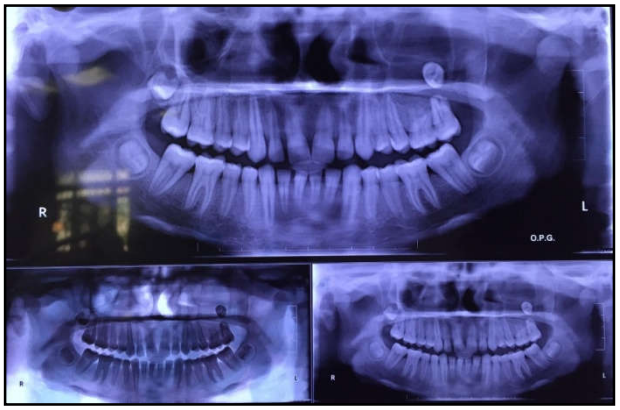


Fig. 2. Orthopantamogram revealed a unilocular radiolucent lesion extending from maxillary left second premolar to maxillary left second molar along with impacted third molar attached inferiorly to roots of teeth and also show resorption root of first and second molar and lesion extending superiorly in the maxillary sinus

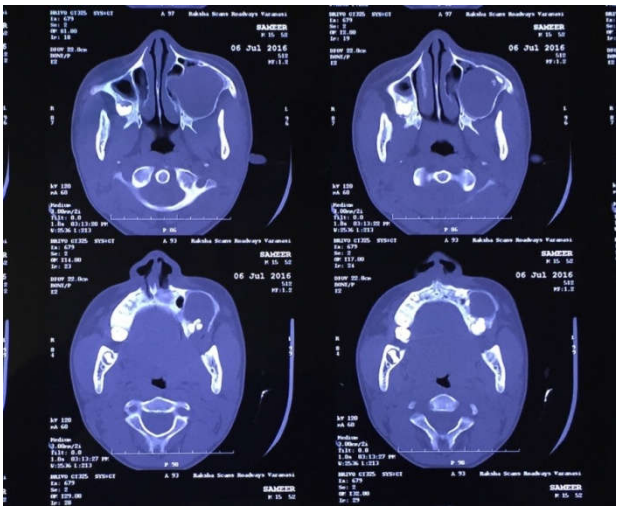


Fig. 3. CT sections revealing monocortical buccal expansion and also expansion of anterior wall of maxillary sinus. CT also revealed haziness and complete obliteration of left maxillary sinus and it also contained an impacted tooth

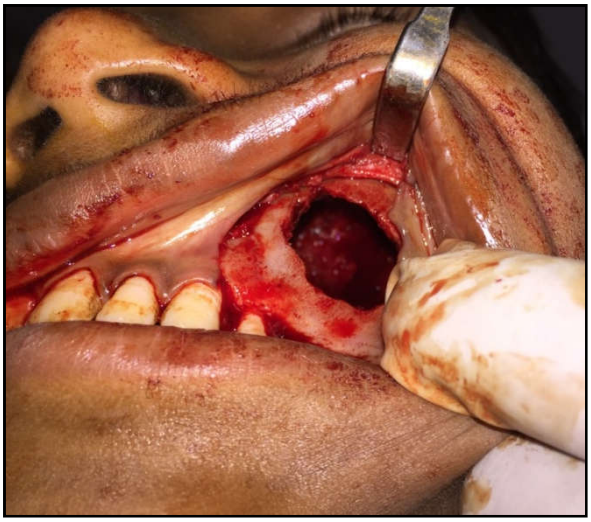


Fig. 4. Intra-operative photograph showing cavity after enucleation

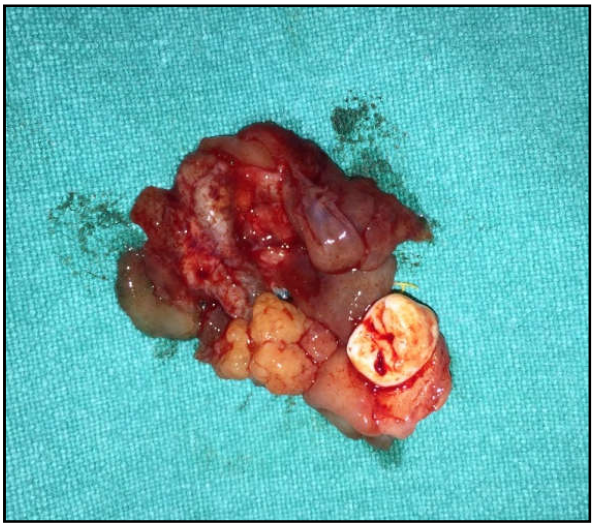
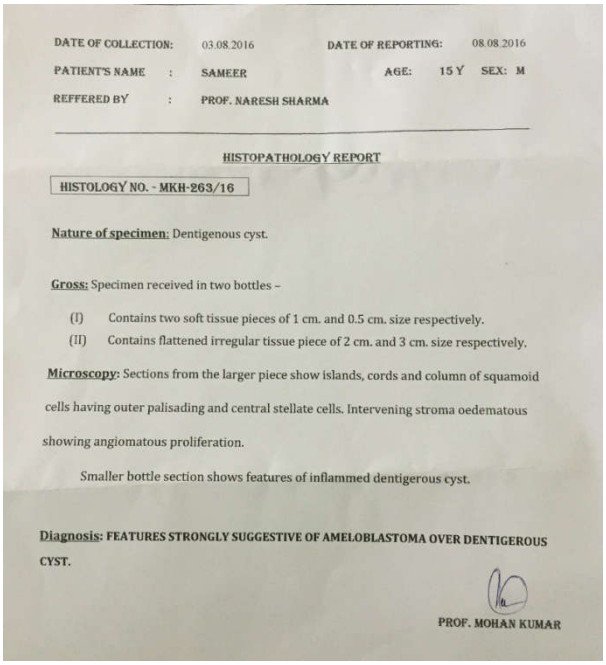


Fig. 5. Specimen showing cystic lesion along with impacted third molar



DATE OF COLLECTION: 03.08.2016 DATE OF REPORTING: 08.08.2016

PATIENT'S NAME : SAMEER AGE: 15 Y SEX: M

REFFERED BY : PROF. NARESH SHARMA

HISTOPATHOLOGY REPORT

HISTOLOGY NO. - MKH-263/16

Nature of specimen: Dentigenous cyst.

Gross: Specimen received in two bottles –

(I) Contains two soft tissue pieces of 1 cm. and 0.5 cm. size respectively.

(II) Contains flattened irregular tissue piece of 2 cm. and 3 cm. size respectively.

Microscopy: Sections from the larger piece show islands, cords and column of squamoid cells having outer palisading and central stellate cells. Intervening stroma oedematous showing angiomatous proliferation.

Smaller bottle section shows features of inflammed dentigerous cyst.

Diagnosis: FEATURES STRONGLY SUGGESTIVE OF AMELOBLASTOMA OVER DENTIGEROUS CYST.

PROF. MOHAN KUMAR

Fig. 6. Histopathological examination revealed ameloblastoma over dentigerous cyst

DISCUSSION

One of the most common cyst that occur in the jaw is dentigerous cyst. Most common presentation of a dentigerous cyst is the occurrence of asymptomatic unilocular radiolucency which encloses the crown of an unerupted or an impacted tooth. The diagnosis of dentigerous cyst is straight forward, but even radiographically, a typical dentigerous cyst can be diagnosed as something else, such as hyperplastic dental follicle, odontogenic keratocyst or a unicystic ameloblastoma on histological diagnosis (Zhang *et al*., 2010). Therefore the diagnosis of these lesions are critical. Dentigerous cyst most commonly seen in the age group of 10-30 years and most commonly associated with an unerupted mandibular 3rd molar in contrast to our case in which lesion associated with maxillary 3rd molar (Deshmane and Khot, 2016). Since dentigerous cyst are often asymptomatic and are usually diagnosed on routine dental radiographs. The diagnosis of a dentigerous cyst is mostly based on a combination of radiographic and histopathological features (Sumer *et al*., 2007). Dentigerous cysts form within lining of dental follicles when fluid accumulates within the follicular epithelium and crown of developing or unerupted tooth (Findic and Huge, 2012). The epithelium of odontogenic cysts can also be transformed into odontogenic tumours like ameloblastoma and adenomatoid odontogenic tumours, or non odontogenic tumours. Various factors that leads to conversion of odontogenic cysts into ameloblastoma includes non specific irritational factors (extraction, trauma, infection, inflammation, unerupteed tooth), nutritional deficiency, viral infections (Ayhan *et al*., 1998). Ameloblastoma is one of the most common types of odontogenic tumour, however, it accounts for only 1% of all oral tumours. It is a slow growing neoplasm, usually occurring in young adults 20-40 years old, with almost equal distribution among men and women. In 80% of patients it occurs in mandible and 70% of these arise from the molar-ramus area (Ayhan *et al*., 1998). Unicystic ameloblastoma is a rare entity.

Those cystic lesion that show clinical , radiographic, or gross features of a jaw cyst but histologically shows atypical ameloblastomatous epithelial lining part of the cyst, with or without luminal and/ or mural tumour growth referred to unicystic ameloblastoma. It accounts for only 6% of all ameloblastomas. It has more occurrence in mandible as compared to maxilla in the ratio 13:1. This tumour is observed in mandibular –ramus region, while posterior region of maxilla is considered rare and atypical (Philipsen and Reichard, 1998). In our case it was associated with the maxillary 3rd molar. It was presented with a painless swelling and facial asymmetry over left side of face. Radiographic evaluation revealed unilocular lesion with well defined sclerotic border. The differential diagnosis of unicystic ameloblastoma should include keratocystic odontogenic tumour, residual cyst, central fibroma, central giant cell granuloma and dysplastic fibrosis. Ackermann *et al*. (1998) and Robinson and Martinez (1997) argued that as epithelium of odontogenic cysts ameloblast omatous share a common ancestory, a transistion from a non neoplastic cyst to a neoplastic could be possible. The minimal criteria for diagnosing a lesion as unicystic ameloblastoma histologically is the presence of cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen in focal areas. Unicystic should be differentiated from odontogenic cysts because the former has a higher rate of reccurence than the latter. In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann (Philipsen and Reichart, 2004) classified unicystic ameloblastoma into the following three histological types:

- ✓ Luminal UA (tumour confined to mthe luminal surface of cyst)
- ✓ Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumour cellsw into the connective tissue wall)
- ✓ Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

Another histologic sub grouping by Philipsen and Reichart has also been described (Konouchi *et al*., 2006)

Subgroup1:Luminal UA
Subgroup1.2:Luminal and intraluminal
Subgroup1.2.3.Luminal,intraluminal and intramural
Subgroup1.3.Luminal and intramural

Unicystic ameloblastoma considered to be less aggressive form that can be successfully removed by simple enucleation or other less aggressive form of surgery. In 1988 Stoelinga and Bronkhorst suggested the use of Carnoy's solution to decrease the risk of recurrence after conservative surgical treatment (Stoelinga and Bronkhorst, 1988). The recurrence rate is generally reported 10-20% and on average less than 25 % after conservative surgical treatment for UA's (Gardner and Corio, 1984). Comparing 50-90% recurrence rates in solid or mulyicystic ameloblastomas after conventional curettage (Ameerally *et al*., 1996). According to Lau and Samman (Lau and Samman, 2006), recurrence rates are 3.6%for resection,30.5% for enucleation alone,16% for enucleation followed by application of Carnoy's solution and 18% by marsupialization followed by enucleation. It was also suggested by one of the study that the vigorous curettage should be avoided because it may implant foci of ameloblastoma more deeply in bone.

Conclusion

Diagnosis of unicystic ameloblastoma preoperatively often pose a difficult situation to the surgeon as it has significant similiarities in clinical and radiographic appearance with odontogenic cyst and tumours so it is important to diagnose lesion histologically as it is one of the most sensitive tool to differentiate dentigerous cyst from ameloblastoma. Since the origin of ameloblastoma is from odontogenic epithelium so it may arise from dentigerous cyst linings as well as from any odontogenic epithelium. Therefore this case report regards dentigerous cyst as a pre ameloblastomatous condition and should be viewed cautiously.

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