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ABSTRACT

Ameloblastoma is a rare, benign odontogenic tumour associated with a high recurrence rate. It accounts for 1% of all tumours of the jaws. The ameloblastoma occurs in three different variants, each with specific implications for treatment and a unique prognosis: solid or multicystic, unicystic, and peripheral. The term unicystic ameloblastoma has been used to describe an ameloblastoma developing within the lining, lumen, or wall of a cyst as well as an invasive ameloblastoma that has a single cystic space rather than multicystic spaces. It accounts for 5-15% of all intraosseous ameloblastomas. Unicystic ameloblastoma has uniquely distinct nature on recurrence as compared to most other variants of ameloblastomas. Lower recurrence rate of unicystic ameloblastoma gives way for conservative treatment rather than the traditional aggressive approach. We report a case of unicystic ameloblastoma of 22 year old female patient.

INTRODUCTION

Ameloblastoma is defined as the “usually unicentric, non-functional, intermittent in growth anatomically benign and clinically persistent” by Robinson. Ameloblastoma is most common benign tumor of jaw. Ameloblastoma is classified as intraosseous, extraosseous & solid or multicystic, unicystic, peripheral. According to the literature amongst all the various types of ameloblastomas the unicystic ameloblastoma has least recurrence rate. Therefore unicystic ameloblastoma are not treated as aggressively as in multicystic ameloblastomas. Generally the cases of unicystic ameloblastomas are seen in 2nd & 3rd decade of life, also when large lesion are aggressively resected to cure the disease, which can cause a psycho-social trauma to the patient in such young age accompanied with loss of function and major deformity. We present a case of unicystic ameloblastoma.

CASE REPORT:

We encountered a patient with swelling on left side of face, her history of multiple operation was an unusual presentation of the recurrent swelling,

which was operated first in 2019 July in which tooth extraction of 38 was done with drain placement and antibiotics were given. In 2020, January again swelling occurred & was treated with the enucleation under L.A and its excisional biopsy diagnosed it as the ameloblastic fibroma. In may 2022, again a swelling was noted on same side of the face for which the CT scan was advised and patient was referred to ADCH for further treatment. According to the history given by the patient the swelling was operated twice in very short period of time and had recurrent swelling for the third time which suggest inadequacy in surgical management of the lesion or reoccurrence. The latest CT scan showed the large radiolucent, unilocular lesion over left ramus region and incisional biopsy was done. It was diagnosed as the unicystic ameloblastoma, as per the diagnosis considering the lower recurrence rates of unicystic ameloblastoma cases & also considering the age of the patient, rather than a aggressive approach we took conservative approach with enucleation with peripheral osteotomy and patient is put on regular follow up.

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Figure. 1 The very 1st opg of patient in July 2019

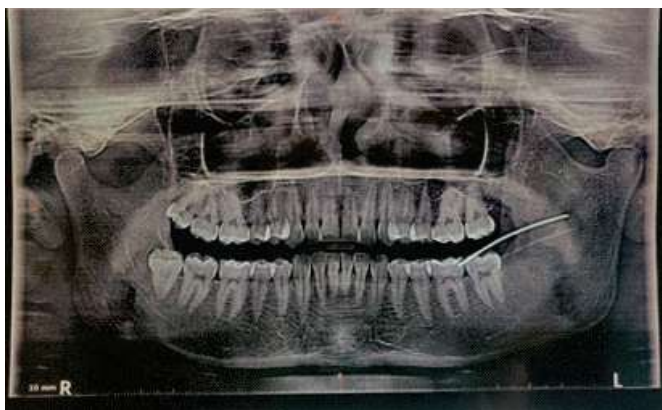


Figure. 2 Extraction of 38, Drain placed in 38 region

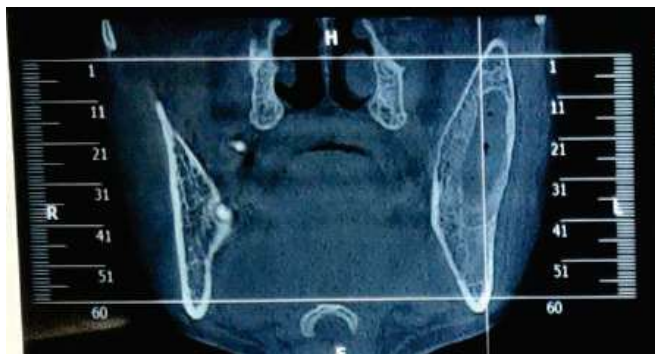


Figure. 3 Cbct in 2020



Figure. 4 Current opg July 2022



Figure. 5 Clinical presentation of swelling at left side of face (2022).



Figure. 6 Intraoral approach was taken for enucleation



Figure. 7 follow up (6 months)



Figure. 8 follow up (6 months)

DISCUSSION

Unilocular ameloblastoma (UA) is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas. Unicystic ameloblastoma were described by Robinson & Martinez. The term unicystic ameloblastoma was adopted in the second edition of the international histologic classification of odontogenic tumors. The term unicystic is derived from the macro and microcystic appearance, this lesion is like a well-defined single cystic sac lined by odontogenic (ameloblastomatous) epithelium.

The ameloblastoma etiology is described in the literature from multiple factors, like it forms from reduced enamel epithelium, it converts into ameloblastoma from a dentigerous cyst also when a solid ameloblastoma undergoes cystic transformation. The age of the patients was 25.5 years with 46% of cases occurring in the second decade and they are most often seen in male patients¹.

In a clinicopathologic study of 57 cases of UA , Ackermann classified this entity into three histologic groups²:

I. luminal UA (tumor confined to the luminal surface of the cyst);

II. Intraluminal/plexiform UA (nodular proliferation into lumen without infiltration of tumor cells into connective tissue wall); and

III. Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

Another subgrouping by Philipsen and Reichart has also been described as follows³:

Subgroup 1: luminal UA;

1.2: luminal and intraluminal;

1.2.3: luminal, intraluminal and intramural; and

1.3: luminal and intramural.

The UA diagnosed as subgroups 1 and 1.2 can be treated conservatively (careful evaluation), whereas subgroups 1.2.3 and 1.3 showing intramural growths require radical resection.

The most common affecting site is the mandibular third molar region, angle region. In this case lesion is seen in the ramus region. The unicystic ameloblastoma has lesser recurrence rate, so treating conservatively is an option. The treatment of ameloblastoma can differ from simple enucleation to aggressive resection. As this case was diagnosed as the unicystic ameloblastoma in incisional biopsy and looking to the recurrence rate, age of the patient treating patient conservatively is treatment of choice. Aggressive resection can cause a psycho-social trauma to the patient in such young age accompanied with loss of function and major deformity, also looking to patient economic condition in which patient is not able to spend on reconstruction. So we took conservative approach and put patient to regular follow ups. Recurrence is also related to the histologic subtypes, among which those invading the fibrous wall have a rate of 35.7%, but others have a rate of only 6.7%⁴. According to Lau and

Samman's systematic review recurrence related to treatment modalities of unicystic ameloblastoma, there is weak evidence showing that resection resulted in the lowest recurrence rate (3.6%), followed by enucleation with application of Carnoy's solution (16%). Enucleation alone resulted in the highest recurrence rate (30.5%)⁵. Unicystic ameloblastomas can be successfully removed by simple enucleation or less aggressive surgery.

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