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Title: A Conservative Approach For Unicystic Ameloblastoma : A Case Series

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Abstract:

The Unicystic ameloblastoma are those cystic lesions which shows a similar features of jaw cyst, but on histopathological examination shows a typical epithelial lining part ameloblastoma . Discovered first in 1977, It is a separate clinicopathological entity than the solid ameloblastoma. It occurs in younger age group with 50% of cases occurring in 2 nd decade. The lesion is not aggressive as a solid ameloblastoma. Histopathological findings is necessary for treatment and prognosis. The management of cystic lesions in young patient varies depends on the clinical behaviour and histopathological diagnosis. It has less recurrence with enucleation than any other ameloblastoma. Hence we present 3 cases of unicystic ameloblastoma in young age group(2^{nd} and 3^{rd} decades of life) treated by enucleation with carnoy's solution and had long term follow up and no recurrence were noted so far in all the cases.

INTRODUCTION-

Unicystic ameloblastoma introduced first in 1997 by ROBINSON AND MARTINEZ. [1] It is a separate clinicopathological entity which resembles like cystic lesion but on the histopathological examination shows a typical ameloblastomatous epithelial lining.[2] Ameloblastoma is the common odontogenic tumour of the jaw. Various studies reported that between 15 to 30% of ameloblastomas occurred from dentigerous cyst.[3] It is 2nd most common odontogenic tumour occurs in a younger age group ,with about 50% of cases seen in the 2nd decade of life. More than 90% are located in the mandible, 50 to 80% of cases with impacted third molar tooth. [4] Incidence of ameloblastoma estimated at 0.5 per million populations per year. But a higher incidence has been reported in several countries like in Africa and Asia.[5]

It may form by enamel organ , reduced enamel epithelium and epithelial cell rests of Malassez. The most common site of occurrence is angle and ramus region of mandible. Clinically lesion is asymptomatic and causes asymmetry of face. [6] OPG shows a unilocular or multilocular radiolucency with root resorption.[7] Ameloblastoma are of two types, Unicystic and solid.[8] Histopathologically characterized as a cystic lesion lined by an ameloblastomatous epithelial lining, and it is classified as luminal, intraluminal and mural sub types.[9]

It often misdiagnosed with dentigerous cyst or odontogenic keratocyst and confirmed diagnosis is done by aspiration and incisional biopsy.[10] Management of unicystic ameloblastoma includes enucleation , curettage and marsupialization where as other treatment modalities are segmental or marginal resection. Conservative approach is preferred in younger patients. [11] Here we report a 3 cases of unicystic ameloblastoma in young patients which were managed by conservative approach , Such as removal of cystic lesions in toto and chemical cauterization with Carnoy's solution.

CASE 1-

A 16 year old male patient visited to our department with facial asymmetry on left side of face . On examination there was diffuse swelling. It was small initially and increases gradually to attend a present size.(figure 1) There was no significant medical and dental history. Clinically swelling present on left body and ramus region of the mandible antero-posteriorly. Superiorly extended from corner of mouth to tragus and inferiorly involving lower border of mandible. Measuring approximately 3x3cm. On palpation swelling was firm and non- tender. Intraoral examination revealed mild diffuse swelling extends from 35 to 37 region. Pus discharge seen between 36 and 37. Grade I mobility noted with 36 and 37. There was obliteration of lower left buccal vestibule. The overlying mucosa was normal. There was no associated pain, difficulty in mouth opening , chewing, and no paraesthesia. Left submandibular lymph nodes are tender on palpation.

OPG shows well-defined unilocular radiolucency with corticated border extending from mesial of 34 to distal of 37. Root resorption and displacement seen with 36 and 37.

(figure 2) Routine investigations were within normal limits. Incisional biopsy was taken and specimen sent for histopathological analysis. And histopathological finding shows the cystic lumen lined by odontogenic epithelial lining which consists of basal layer of cuboidal to columnar cells. The basal cells are hyperchromatic and shows reversal of polarity. The connective tissue capsule is fibrocellular consisting of parallelly arranged collagen fibre bundles interspersed with fibroblast. Diffuse intense chronic inflammatory cell infiltrate is seen subepithelially mainly in the form of lymphocytes and plasma cells. Endothelial lined vascular channels with extravasated and intravasated blood elements are also seen. These overall features are suggested of luminal unicystic ameloblastoma.

Considering the patients age, size of lesion and histologically confirmed diagnosis of Subgroup 1 as a luminal unicystic ameloblastoma. Conservative treatment plan was decided under local anaesthesia that is removal of entire cystic lesion and chemical cauterization with Carnoy's solution.(Figure 3) Excised tissue was sent for histopathological examination and patient was given I.V antibiotics and analgesics for a week. Ryle's tube feeding was given for a week and patient advised liquid diet. Patient kept for follow up once in 15 days for 6 month and then once in month for 3 years . The healing was satisfactory.

FIG.1--Preoperative extraoral view.

Fig.2 - OPG showing cystic lesion.

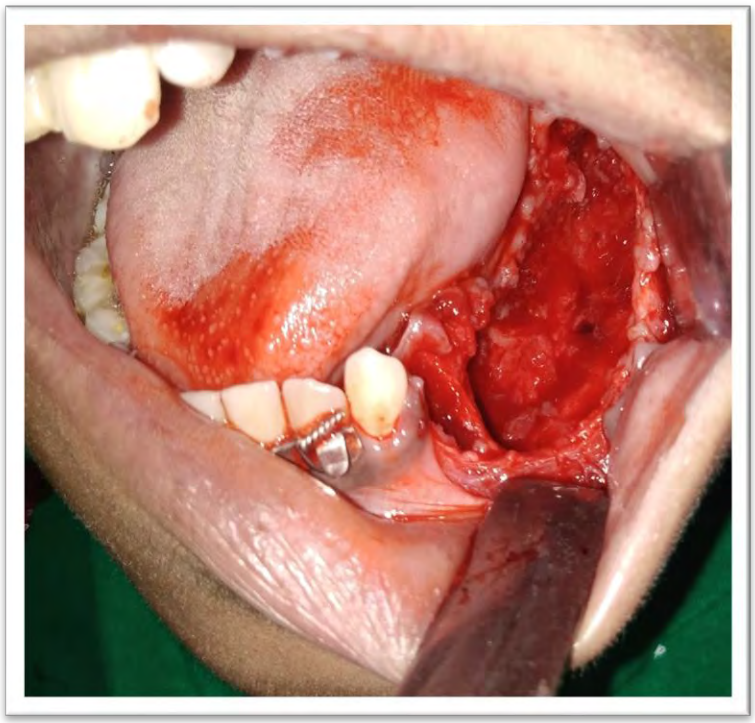

Fig 3-Enucleated cystic lesion.

CASE 2-

A 17 year old female patient complains of swelling on the left side of face. On extraoral examination swelling presents on the body and ramus region of the left side of mandible. (figure 4) Swelling was firm and non-tender in nature. There was no history of pain, toothache, pus discharge and paraesthesia. No neck nodes were palpable. Intraorally diffuse swelling extends from the distal aspect of 35 till the pterygomandibular region. Vestibular obliteration was present. Grossly carious with 36 , grade I mobility with 35 noted. OPG shows well defined unilocular radiolucency in the left body region extending from mesial of 36 to the ramus till the left sigmoid notch and 37 was displaced towards coronoid process. (figure 5) CBCT of mandible was done , Axial view showed 5.6x 3cm unilocular radiolucent lesion with expansion of both the cortical plates and perforation on both sides was seen. (figure 6)

Histopathological examination of incisional biopsy was suggestive of unicystic ameloblastoma. Considering the patients age, size of lesion and histologically confirmed diagnosis of Subgroup 1 as a luminal unicystic ameloblastoma , conservative treatment plan was decided. That was removal of cystic lesion in toto followed by chemical cauterization with carnoy's solution. Patient was kept under observation. Intravenous antibiotics and

analgesics were given for a week. And patient was kept on Ryle's tube, on soft and liquid diet for 1 week. Follow up was done . The healing was satisfactory.

Fig.4-Preoperative extraoral view.

Fig.5- OPG showing cystic lesion

Fig.6 -CT scan -preoperative showing cystic lesion in left side of mandible.

CASE 3-

A 26-year-old male reported with complain of painless swelling with lower left back region of jaw since 3 years. The swelling was gradually increased to present size. (Figure 7) Past medical and dental history was not relevant. Clinical examination revealed swelling present on left body and ramus region of the mandible with diffuse borders. On palpation, swelling was firm and non-tender in nature. Intraoral examination showed a swelling from 34 to 38 with obliteration of vestibule. Root stump present with 36 and deep occlusal caries seen with 46. The overlying mucosa was normal.

OPG showed well-defined unilocular radiolucency extending from distal of 33 to distal of 38. Root resorption with 37 was visible .(Figure 8) Axial view showed 4.3x 2cm radiolucent lesion with expansion of both cortical plates. (Figure 9) The routine general investigations was within normal limits and incisional biopsy was suggestive of unicystic ameloblastoma and hence the cystic lesion was removed in toto with extraction of 35,36 and 37 and chemical cauterization with Carnoy's solution was done. (Figure 10) Initially follow up was done once in 15 days for 6 months, then patient was called once in a month. The healing was satisfactory and no recurrence noted so far.

Fig.7- Preoperative extraoral view.

Fig.8- OPG showing cystic lesion.

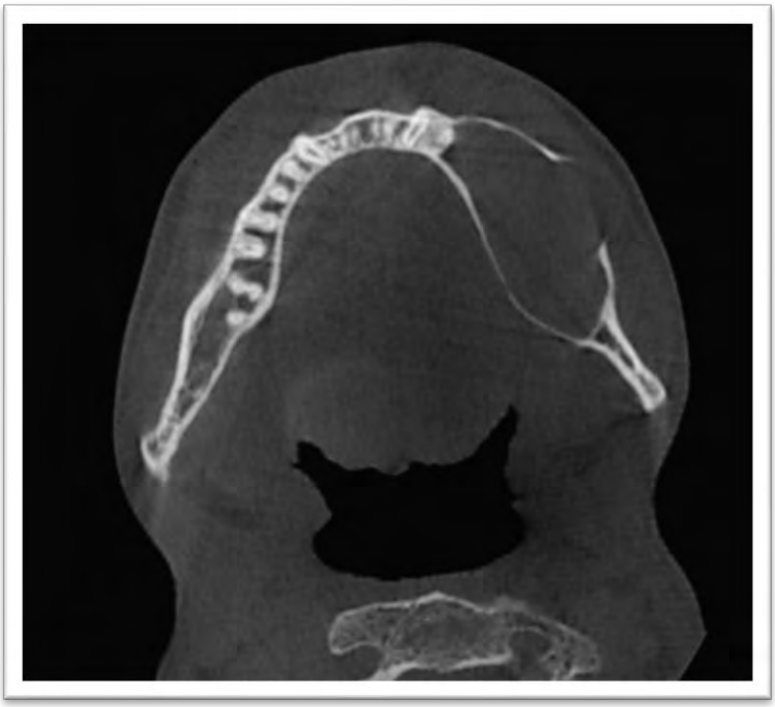

Fig.9- CT scan -preoperative showing cystic lesion.

Fig.10-Exicised specimen.

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DISCUSSION-

Ameloblastoma is the common tumour of odontogenic origin which is locally invasive or aggressive but having benign characteristics. Its accounting for approximately 1% of all odontogenic tumours.[6, 7] There are three forms of ameloblastoma unicystic, multicystic and peripheral. [8] Ameloblastoma has an unique biological behaviour of slow growing ,locally invasiveness and high recurrence rate. [13] WHO defined it in 1992 as a benign but locally invasive neoplasm consisting of odontogenic epithelium, which is either follicular or plexiform pattern.[14] Unicystic ameloblastoma is a separate clinicopathologic entity which resemblance like a cystic lesion but on histopathological examination it shows a typical ameloblastomatous epithelial lining part of the cyst cavity.[2] It is reported that between 15 to 30% of ameloblastomas occurred from dentigerous cyst.

23.8 years of mean age has been reported for unicystic ameloblastoma by Ackerman et al. The male/female ratio of 1.3 :1 as per study of Ademola et al agrees with the findings of Ackerman et al who also found a same ratio.[15] UA is the most common type of ameloblastoma in younger than 20 yrs old patients mostly in Caucasian patients and also more common in western countries then in Africa and Asia . In underdeveloped countries , patient admit themselves to hospital only when they have incapacitating symptoms . In contrast , in developed countries ameloblastoma are frequently detected during routine dental check -ups.[5]

The most common site of unicystic ameloblastoma seen in molar and ramus region of the mandible associated with tooth impaction. [2] In our cases the lesion were asymptomatic , patients commonly present with facial asymmetry and painless swelling of the jaws. Pain is an occasional sign, painless swelling often indicates a lesion of long duration and significant size. Radiographically unicystic ameloblastoma presents primarily as unilocular

radiolucency but may occasionally exhibit multilocular radiolucency.[7] It commonly occurs as a peri-coronal radiolucency resembling a dentigerous cyst. The unicystic indicates a unilocular appearance , the lesion can rarely have a multilocular appearance.[6] Unilocular are more common than multilocular unicystic ameloblastoma with an impacted tooth.[12] Unilocular pattern is often misdiagnosed as an odontogenic keratocyst or a dentigerous cyst and is seen in cases associated with tooth impaction.[2]

Microscopically the lesion appears as a well- defined , often large mono-cystic cavity with a lining , focally but entirely composed of odontogenic epithelium. It is often accompanied by not harmful epithelium of varying histological appearance that may mimic the lining of a dentigerous or radicular cyst.[6] Histologic group classified by Ackermann et.al are[4]

Group 1-Luminal type (tumour confined to the luminal surface of the cyst)

Group2-Intraluminal/Plexiform (cystic lesion comprising intraluminal tumour nodules and odontogenic epithelium with a plexiform pattern, resembles the one seen in plexiform solid ameloblastoma)

Group 3-Mural type (With the presence of ameloblastomatous epithelium tumour islands in the cyst wall, which may or may not attached to cystic lining.)

Another histologic subgrouping by Philipsen and Riechart has also been described –[4]

Subgroup 1-Luminal unicystic ameloblastoma

Subgroup 1.2-Luminal and intraluminal

Subgroup 1.2.3- Luminal, intraluminal and intramural

Subgroup 1.3-Luminal and intramural

Histopathological diagnosis , being a gold standard , clinicians are usually guided by a treatment plan based on initial histopathological findings even when it differs from the clinical diagnosis.[12]

Treatment planning depends on the patients age , tumour size, location, radiographic appearance, final histopathological diagnosis. Enucleation and curettage is the most acceptable treatment modality for luminal and intra-luminal unicystic ameloblastoma . For Mural type a radical approach and the same had been stated by Black et al , Kessler HP and Gardner .[7] In our cases we followed a conservative treatment modalities considering a patient age.

The use of carnoy's solution was initially proposed by Stoelinga and bronkhrost in 1987. And recently proposed again as a possible means to diminish the recurrence risk after conservative treatment. Cornoys solution (chloroform 3 ml , absolute alcohol 6 ml, glacial acetic acid 1 ml and ferric chloride 1 g) was described in 1933 as a sclerosing agent for treatment of cyst and fistulae and remains in use today as fixative. Carnoy's solution had been shown to penetrate the cancellous bone.[1] Rosenstein et al and Lee et al reported success rates with recurrence rates of 10% by using carnoy's solution as an adjunct to enucleation and curettage.[3]

Some studies have evaluated more conservative therapies with good results and where it is suggested to start with this modality before performing more aggressive surgery. Huang et al conducted a study in 15 cases of

ameloblastoma with age of under 18 years and concluded that good results can be obtained with conservative surgery and that in case of recurrence , a second surgery can be successful.[9] Tanaka et al reported in his study that first approach should be minimal surgical treatment in unicystic ameloblastoma in children's. Treatment is complicated because of three factors -1)The continuing facial growth and different bone physiology . 2)unerupted teeth. 3)The difficulty in initial diagnosis.[10] And hence in our cases we prefer the conservative approach to the unicystic ameloblastoma.

In our cases attempt was made to treat all the cases by conservative treatment (surgical enucleation and curettage followed by chemical cauterization with carnoy's solution.) Our all 3 cases had a younger age group and responded well to a conservative management and no recurrence has been reported so far in all cases .

CONCLUSION-

The diagnosis of unicystic ameloblastoma based on clinical features, radiographic features and histopathological examination . Although the management of UA remains controversial, Conservative approach is preferable in younger age group to offers better quality of life. In our cases patient was treated by a conservative approach and did not have any complications and recurrence so far.

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BDS in 2001 and MDS in Oral and Maxillofacial Surgery (Master of dental surgery) in 2006. Joined as a lecturer in MGM Dental College Navi Mumbai in 2006-2007, Then joined as a lecturer in GDC Mumbai in 2007-2013. Presently as Associate professor in GDC Mumbai since 2013-till date. Got a Best Paper Award in 2005 for paper -Hoping for a Blissful Goodnight's sleep.(Management of obstructive sleep apnoea). Published 6 papers in international journals and 9 papers in national journals . Have a current interest in oral and maxillofacial Oncology.

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