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MURAL UNICYSTIC AMELOBLASTOMA – A CASE REPORT WITH 7 YEAR FOLLOW-UP

Maxillofacial Surgery

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ABSTRACT

Mural unicystic ameloblastoma is a distinct entity characterized by a solitary cystic cavity lined with odontogenic epithelium which exhibits invasive growth patterns as evidenced by infiltration of ameloblastomatous epithelial cells into adjacent connective tissue wall. We report a case of mural unicystic ameloblastoma located in the right posterior region of the mandible, which was managed with a conservative approach and carefully followed up for a period of 7 years.

KEYWORDS

mural unicystic ameloblastoma, enucleation, carnoy's solution, long term follow up

INTRODUCTION :

The mandible can be affected by various benign tumors, which can be broadly classified into two categories: odontogenic and nonodontogenic tumors. Among the odontogenic tumors, ameloblastoma is the most common tumor, arising from the epithelial cells and dental tissues during different stages of tooth formation. Its behavior is marked by a slow-paced progression, and aggressive properties, including localized tissue penetration, a high likelihood of recurrence, and the ability to metastasize.

The unicystic ameloblastoma is a relatively rare variant of ameloblastoma, characterized by cystic lesions that exhibit clinical and radiographic features reminiscent of odontogenic cysts. However, upon histological examination, these lesions are distinguished by the presence of a typical ameloblastomatous epithelial lining, which may be limited to a portion of the cyst cavity, and may or may not be accompanied by luminal and/or mural tumor proliferation. In this paper we report a case of mural unicystic ameloblastoma located in the right posterior region of the mandible, which was managed with a conservative approach and carefully followed up for a period of 7 years.

Case Report :

A 11-year-old female patient presented to our department with the chief complaint of swelling and loosening of teeth in the lower right posterior region since 1 year and the swelling which had been progressively increasing in size over the past year.

Extraoral examination revealed a diffuse, oval-shaped swelling measuring approximately 8cm x 6cm in size, extending from 3cm from the corner of the mouth to 1cm from the tragus of the ear in an anteroposterior direction, and from 3.5cm below the ala-tragus line to the inferior border of the mandible in a superoinferior direction. The swelling was hard and non-tender to palpation, with no associated skin changes or increase in local temperature.

Intraoral examination revealed a single diffuse swelling in the right buccal vestibule of the mandible, corresponding to teeth 44, 45, and 46. The swelling measured approximately 3x2cm in size and extended from the distal aspect of tooth 43 to tooth 46 in an anteroposterior direction, and from the attached gingiva to the buccal vestibule in a superoinferior direction. Expansion of the buccal and lingual cortical plates was observed in relation to teeth 44, 45, and 46. The swelling was firm in consistency, non-fluctuant, and non-reducible, with no discharge or pulsations noted. The second permanent molar (tooth 47) was found to be unerupted. All teeth in the affected region exhibited grade III mobility. Needle aspiration yielded a brown-yellow fluid.

The CT scan demonstrated a well-defined expansile cystic lesion in the body of the mandible. There was thinning of both lingual and buccal cortical plates, as well as the basal bone.

A preoperative diagnosis of an inflammatory dentigerous cyst and unicystic ameloblastoma was made based on the patient's age, lesion location, and clinical and radiographic features. The patient's parents were counseled regarding the complexity of the diagnosis, treatment options, and the risk of recurrence. Given the patient's age and the complexity of the diagnosis, a comprehensive treatment plan was devised, involving cystic enucleation, application of Carnoy's solution, and extraction of adjacent teeth, to be performed under general anesthesia.

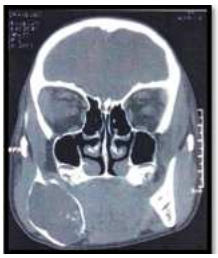
Operative Procedure - Under general anesthesia, a crevicular incision was made extending from tooth 44 to tooth 46, and a mucoperiosteal flap was elevated to expose the underlying lesion. Teeth involved, 44, 45, and 46 were extracted, and the lesion was enucleated in toto, along with the impacted second molar (tooth 47). The bony cavity was thoroughly irrigated with a betadine solution and normal saline to remove any remaining debris. Subsequently, Carnoy's solution was applied to the bony cavity for 3 minutes, followed by rinsing with normal saline. The surgical site was then closed using 3-0 vicryl sutures. The excised lesion was sent for histopathological examination.

Microscopic analysis of the surgical specimen revealed a cystic lesion surrounded by a fibrous tissue capsule, lined by odontogenic epithelium of variable thickness. The epithelial lining exhibited nuclear palisading along the margins and loose stellate reticulum. The cells contained moderate to abundant pale acidophilic vacuolated cytoplasm and round to oval vesicular nuclei. Focal epithelial invaginations were observed, associated with desmoplastic fibrosis.

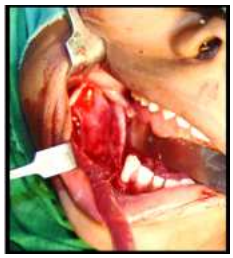
Based upon the radiological and histopathological report, the case was diagnosed as mural unicystic ameloblastoma.

In the subsequent visit removable partial denture was given. Notably, no signs of paresthesia or numbness were observed in the lip or cheek. Given the high recurrence rate of mural unicystic ameloblastoma, a long-term follow-up period was planned to monitor the patient's progress and detect any potential recurrences or complications.

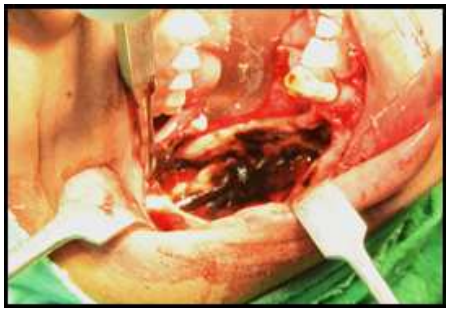
The patient was instructed to return for follow-up appointments at 1 week, then monthly for 3 months, and subsequently every 6 months. Radiographic evaluations were performed at 6-month intervals to assess for any signs of recurrence. Fortunately, no recurrence was observed during the follow-up period



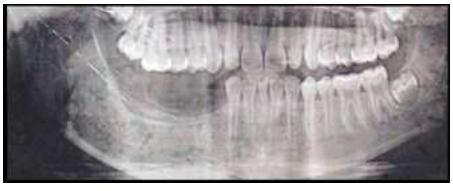
Pre-operative CT coronal view image



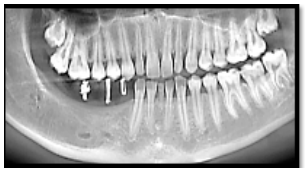
Application of Carnoy's solution



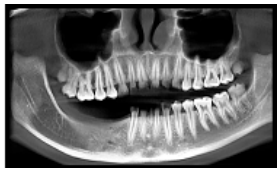
Application of Carnoy's solution



3 Year Follow-up Radiograph



5 year follow up radiograph



7 year follow up radiograph

DISCUSSION :

Ameloblastoma is a slow-growing, benign tumor that originates from odontogenic epithelial cells, primarily derived from undifferentiated enamel tissue. Despite its slow growth rate, this tumor can lead to significant local tissue damage due to its invasive nature. The 2017 edition of the World Health Organization's (WHO) Classification of Head and Neck Tumors categorizes ameloblastomas into four distinct subtypes. These include: conventional ameloblastoma, unicystic ameloblastoma, extraosseous or peripheral ameloblastoma, and metastasizing ameloblastoma.[1] First described by Robinson and Martinez in 1977, unicystic ameloblastoma (UA) is a unique variant of ameloblastoma. Characterized by clinical and radiologic features resembling an odontogenic cyst, UA is distinguished by the presence of a typical ameloblastomatous epithelium lining part of the cyst cavity upon histological examination. This epithelium may exhibit luminal and/or mural tumor proliferation, with or without additional features.[2]

The unicystic variant of ameloblastoma presents most commonly in the pediatric age group.[4] The age of the patient reported is 11 years.

The location within the jaw bones favours greatly the mandible (mandible:maxilla = 3 to 13:1) and the posterior mandible is the most often a affected single region.[3] The region involved in this case is right posterior mandibular region (irt 44, 45, 46 teeth).

On radiographic examination, unicystic ameloblastomas typically present as solitary, expansile radiolucent lesions with a well-defined margin. Notably, a significant proportion of cases, ranging from 50% to 80%, are found to be associated with a tooth that is unerupted or is impacted.[3] Similarly, in our case, there was a solitary expansile cystic lesion with an unerupted permanent mandibular second molar.

The histological analysis of this case revealed the typical morphological features of unicystic ameloblastoma, aligning with the findings of numerous case reports. The histopathological samples showed cystic spaces lined by a flattened epithelial layer with ameloblastomatous characteristics, which exhibited proliferative activity both within the lining and in the surrounding connective tissue. This proliferation resulted in the formation of distinct ameloblastomatous islands, a key diagnostic feature of mural variant of unicystic ameloblastoma.

The management of unicystic ameloblastomas involves a range of surgical approaches, including both radical and conservative methods. Treatment options may include enucleation, marsupialization, chemical cauterization, curettage, radiation therapy, or a combination of surgery and radiation therapy, depending on the specific case and clinical presentation.[5-7]

For unicystic ameloblastomas, located in the mandible, especially for younger patients ,conservative treatments are typically favored. While more aggressive interventions, such as resection, can effectively eliminate the risk of recurrence, they are generally not considered the most desirable treatment option for unicystic ameloblastomas, as they may result in significant morbidity and functional impairment and disturb their psychosocial wellbeing.[8-10]

According to Porgrel et al.[11], the recommended treatment protocol for unicystic ameloblastomas consists of enucleation followed by curettage and adjunctive therapy with either liquid nitrogen cryospray or Carnoy's solution cauterization. The same protocol was followed in this case.

The recurrence rate of unicystic ameloblastoma after treatment varies depending on the approach used. The overall recurrence rate ranges from 10% to 25%. A comparison of the recurrence rates for different treatments is presented below:

Recurrence Rates of Unicystic Ameloblastoma by Treatment [12]

- Resection: 3.6%
- Enucleation alone: 30.5%
- Enucleation with Carnoy's solution: 16%
- Marsupialization (with or without additional treatment): 18%
- Conservative treatment: 55-90%
- Radical approach: 15-25%

These findings highlight the importance of selecting an appropriate treatment strategy and long term follow up is required to minimize the risk of recurrence in patients with unicystic ameloblastoma.[12]

So, after treatment, the patient was monitored for 7 years, with no evidence of recurrence noted during this period.

CONCLUSION :

In adult patients, radical surgical excision is often the preferred treatment approach for mural unicystic ameloblastoma (UA) to minimize the risk of recurrence. However, in pediatric patients, this aggressive surgical approach may compromise craniofacial development, leading to functional and aesthetic impairments that can significantly impact quality of life. Therefore, a more conservative surgical strategy may be warranted in younger patients, with the goal of preserving structural and functional integrity while promoting uneventful healing and bone regeneration. One promising treatment modality for UA in pediatric patients involves enucleation of the lesion followed by application of Carnoy's solution, which has been shown to be effective in reducing tumor recurrence.

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