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UNICYSTIC AMELOBLASTOMA OF THE MANDIBLE - A CASE REPORT

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Abstract :

Ameloblastoma is a true neoplasm of odontogenic epithelial origin, with unicystic ameloblastoma being a subtype that comprises 5-15% of all intraosseous ameloblastomas. It's interesting to see its clinical and histological characteristics manifesting as a cystic lesion resembling a mandibular cyst but revealing ameloblastomatous epithelium upon examination. This case is of a 17-year-old male who adds to the body of literature on this condition.

The fact that it's the second most common odontogenic neoplasm after odontoma highlights its clinical significance. Management of such cases often involves careful surgical intervention due to its tendency for recurrence and potential aggressive behavior.

Understanding the nuances of different subtypes of ameloblastoma, like the unicystic variant, is crucial for accurate diagnosis and appropriate management. Such reports contribute significantly to the understanding and management of this condition.

Introduction :

Numerous benign lesions can cause mandibular swellings, categorized into odontogenic and non-odontogenic origins. These include ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumor, central giant cell granuloma, fibro-osseous lesions, and osteomas. Ameloblastoma is the most common odontogenic tumor, originating from epithelial cellular elements and dental tissues. It is a slow-growing, locally aggressive neoplasm typically occurring in the 3rd to 4th decades of life, with an equal sex distribution, often associated with an unerupted third molar. Routine radiography may detect it.

Most ameloblastomas arise in the mandible, particularly in the angle and ramus region. They exist in three forms: multicystic, peripheral, and unicystic tumors. Multicystic ameloblastoma is the most common, accounting for 86% of cases. Peripheral tumors are odontogenic tumors with histological characteristics resembling intraosseous ameloblastoma but found solely in the soft tissues covering the tooth-bearing parts of the jaws. Unicystic tumors include those referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts.

The primary goal in treating ameloblastoma is complete excision with appropriate reconstruction. We present a case of a large unicystic mandibular ameloblastoma in a 17-year-old male.

Case Report :

A 17-year-old male presented with a gradually enlarging swelling on the right front and back tooth region which gradually increased in size over a period of 2 months (Figure 1.). He reported no associated pain, difficulty in mouth opening, chewing, or speaking. On extraoral examination (Figure 1.2) a diffuse swelling was seen on lower anterior mandibular region. Intraoral examination revealed very slight diffuse swelling along with marked bucco lingual cortial plate expansion extending anterioposteriorly from in lower anterior mandibular extending from left canine to right first molar showing no facial asymmetry. Neither discharge nor pulsations were seen. On palpation the swelling was tender and crepitus was felt on lingual cortical plate. The swelling was firm in consistency and surface was smooth with no rise in temperature. It was non fluctuant and nonreducible. Crepitus could be felt over the swelling. Oral mucosa appeared normal, and no palpable neck nodes were detected.

Radiographic Examination :

An orthopantomogram (OPG) revealed a large unilocular radiolucency large (approximately 3.5 cm × 6.5cm) with well circumscribed radiopaque border extending from 33 tooth region to 46 tooth region. (Figure 2). CT scan confirmed the cystic lesion confined within the mandible with cortical thinning .

Differential Diagnosis :

Our case must be distinguished from other multilocular lesions including solid ameloblastoma, central giant cell granuloma, odontogenic myxoma, as well as cystic lesions like odontogenic keratocyst (OKC), dentigerous cyst (DC), and residual cyst.

Investigations :

Routine investigations like complete blood picture were done, which were normal. Systemic examination revealed no abnormalities. Incisional biopsy was done and submitted for histopathological examination. The histopathological examination of the resected specimen confirmed the presence of a unilocular ameloblastoma with no signs of malignancy.(Figure 3)

Microscopically, a cystic cavity lined by epithelium was observed, with columnar, hyperchromatic, and palisaded basal cells exhibiting reverse polarity. Epithelial proliferation into the lumen, resembling the stellate reticulum, and foci of squamous metaplasia were also noted. Underneath the fibrous capsule, there was proliferation of neoplastic cells arranged in strands or islands, along with areas of squamous metaplasia and islands of odontogenic epithelium. The final diagnosis, based on clinical and microscopic features, was unicystic ameloblastoma.(Figure 4)

Treatment :

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Under strict aseptic conditions, general anesthesia was administered via naso-endotracheal intubation. The patient's face and oral cavity were prepped with povidone-iodine and draped. Local infiltration was done using local anesthesia with adrenaline 1:80,000. An crevicular incision was made from tooth 46 to 33, followed by supraperiosteal dissection over the anterior mandibular region to expose the tumor. The entire lesion was enucleated followed by curettage and chemical cauterization with Carnoy's solution were performed at the lesion site, and a thorough intraoral irrigation. Primary closure was achieved using 3-0 vicryl. Patient was extubated and shifted to the postoperative ward without complications. The patient underwent surgical enucleation with chemical cauterization using Carnoy's solution under general anesthesia. (Figure 5 and Figure 6) .Follow-up for one year post-treatment showed signs of new bone formation on the OPGs taken at 3months interval with no recurrence noted to date (Figure 7).

Figure 1.2 Extraoral Preoperative Photograph

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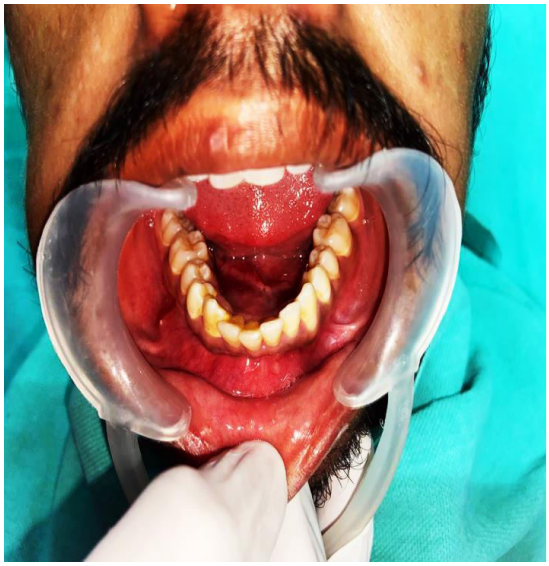

Figure 1.1 Intra Oral Preoperative Photograph

Figure 2 Pre operative Orthopantomogram

Figure 3 Incisional Biopsy was performed

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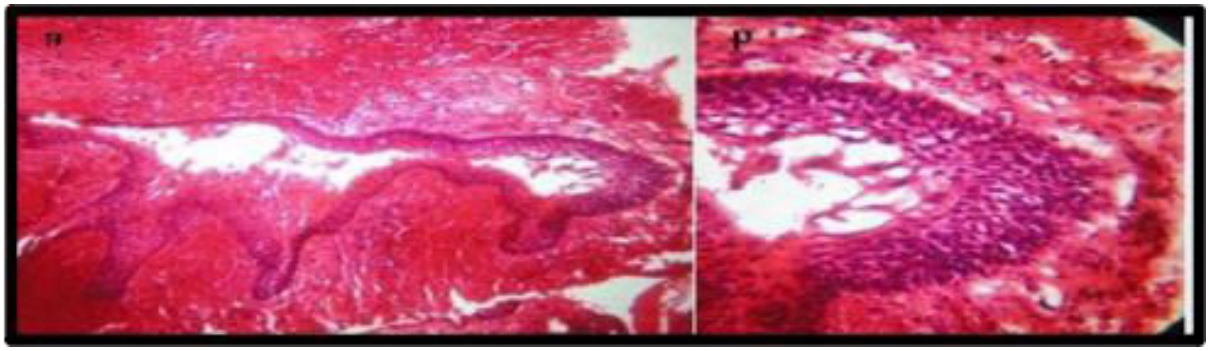

Figure 4 Histopthalogical Examination under Microscope

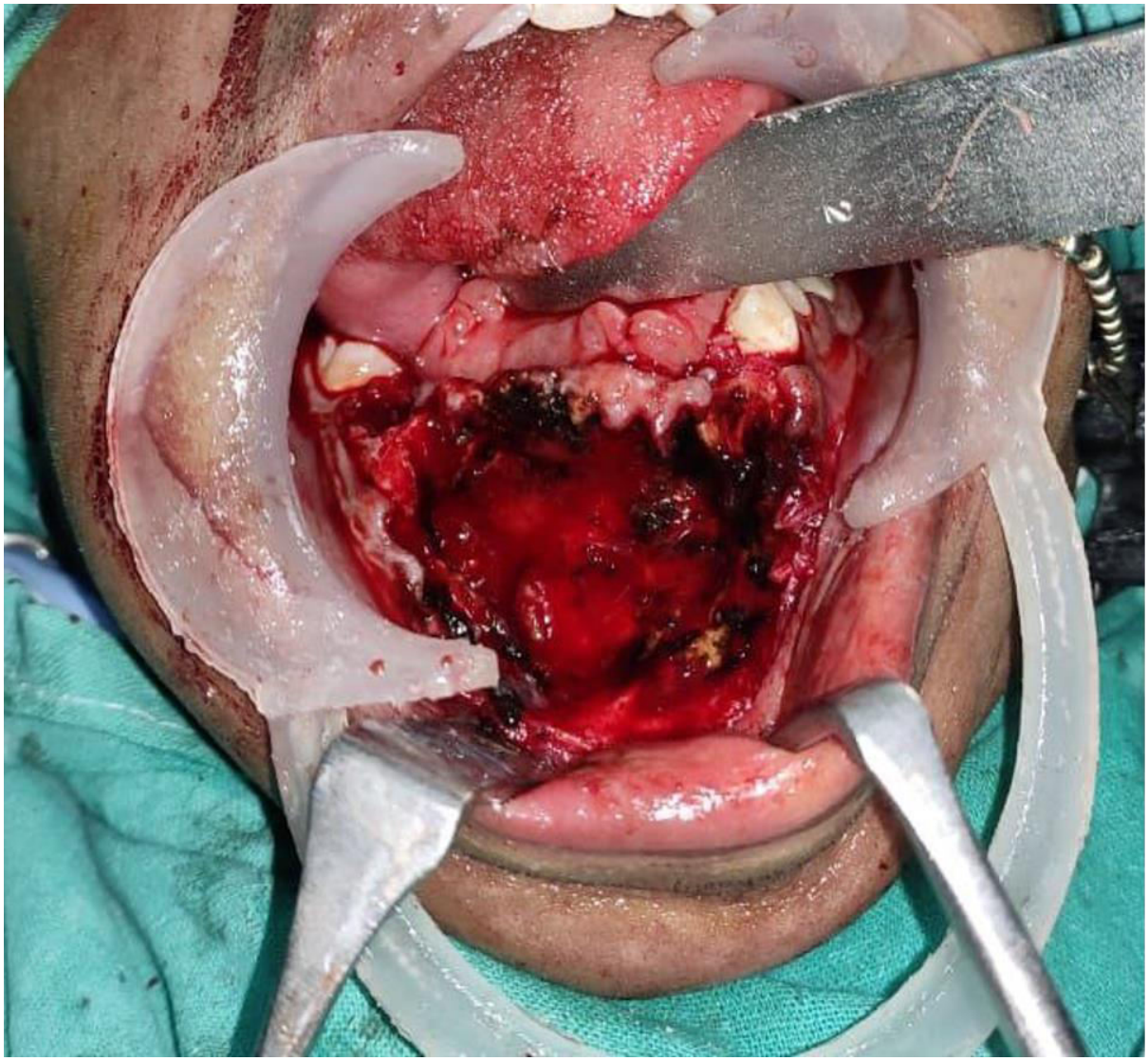

Figure 5 Enucleation of the lesion followed by curettage

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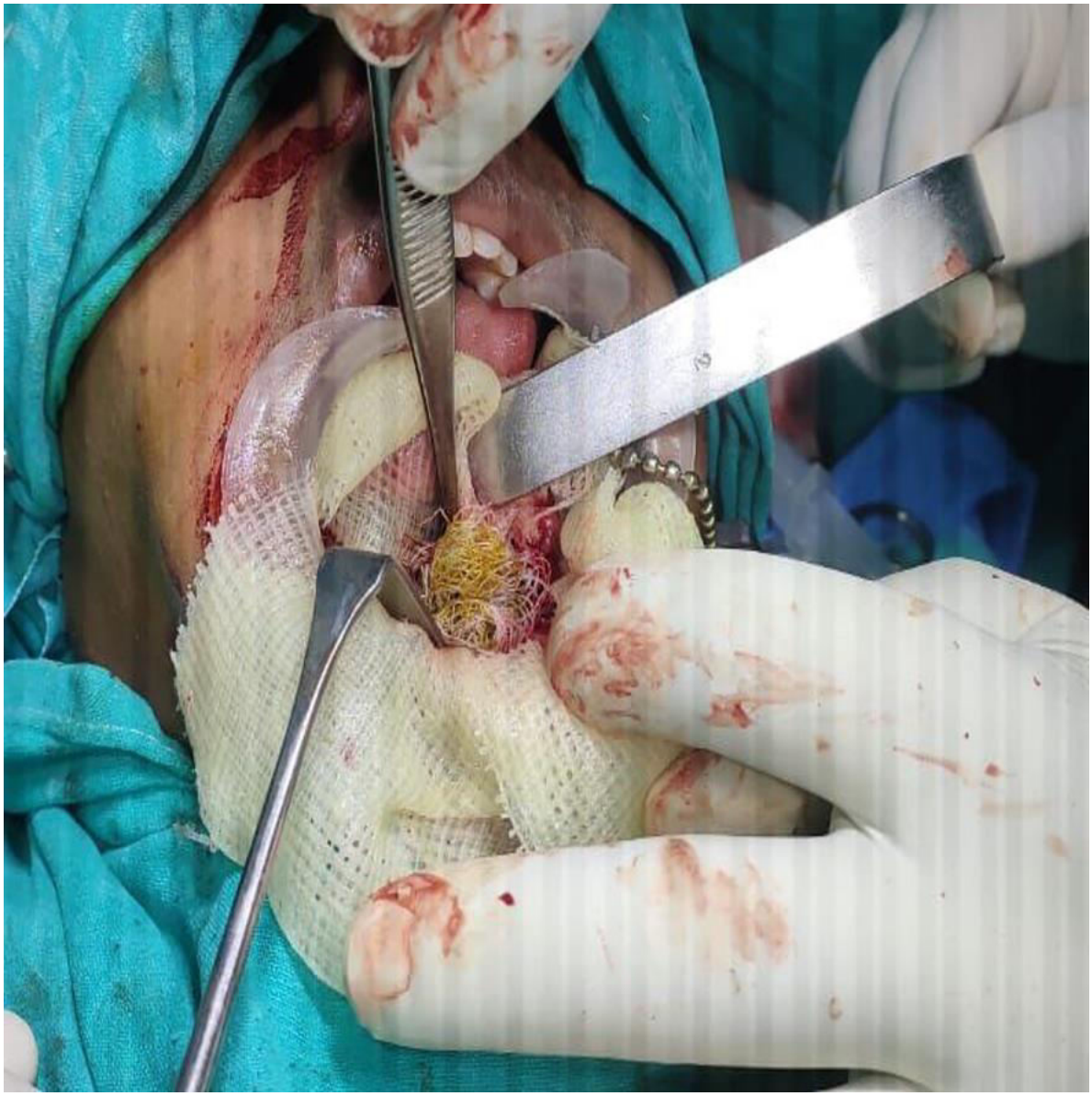

Figure 6 Chemical cauterization using Carnoy's solution

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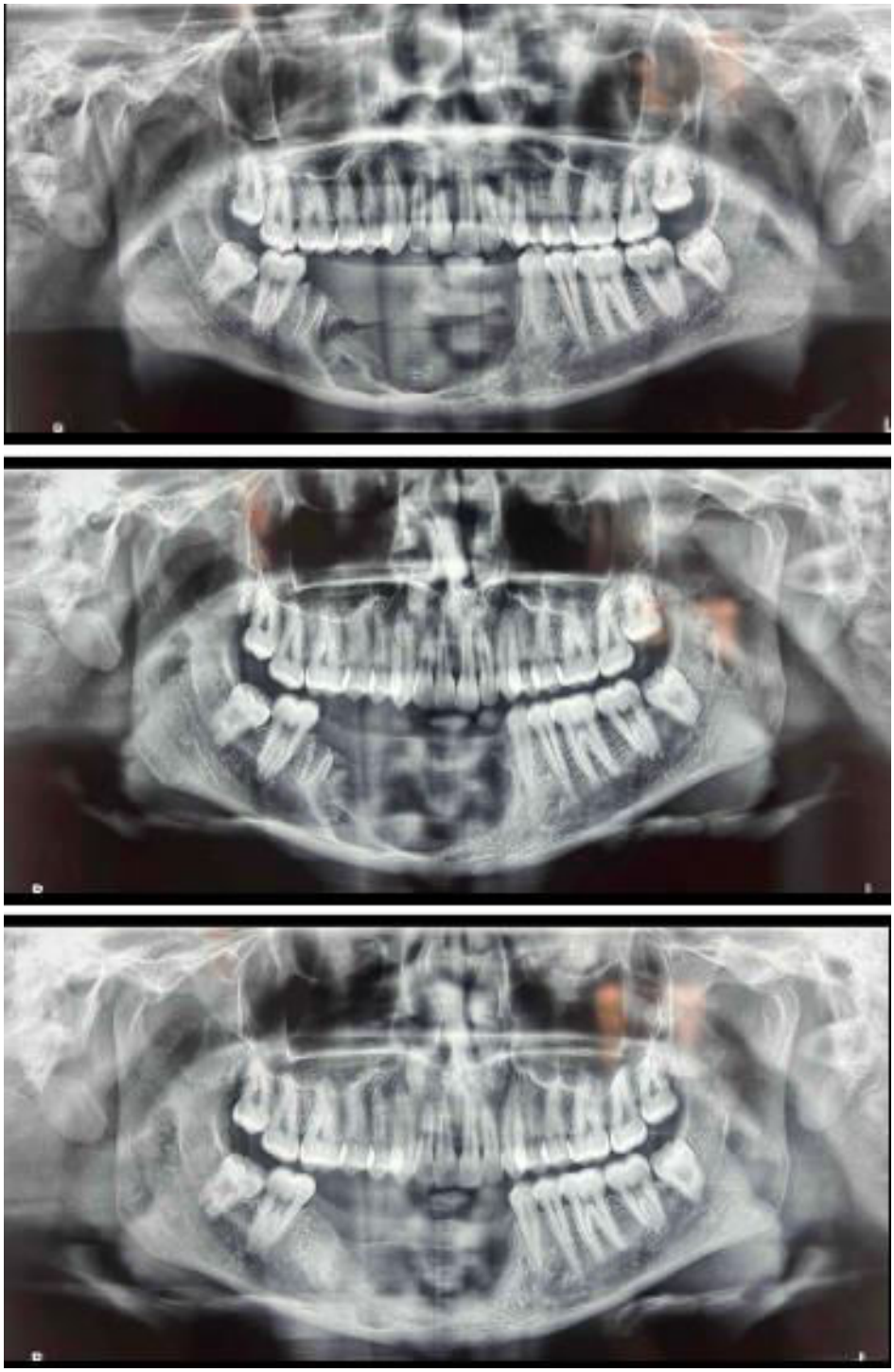

Figure 7 Followup Orthopantomograms at 3 months interval

Discussion :

Unilocular ameloblastoma (UA) is a rare subtype, comprising approximately 6% of all ameloblastomas. It typically affects a younger age group, with around 50% of cases occurring in the second decade of life. More than 90% of UA cases are found in the mandible. Between 50 and 80% of cases are associated with tooth impaction, with the mandibular third molar being most commonly affected. The 'dentigerous' type tends to occur about 8 years earlier on average than the 'non-dentigerous' variant.

Patients commonly present with swelling and facial asymmetry, with pain occasionally being a symptom. Mucosal ulceration is rare but may occur due to the continued growth of the tumor. Small lesions may be detected incidentally on routine radiographic screening or due to local effects such as tooth mobility, occlusal alterations, and failure of tooth eruption caused by the tumor.

Histologically, the minimum criterion for diagnosing UA is the presence of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium, often observed in focal areas. It's important to differentiate UA from odontogenic cysts because UA has a higher recurrence rate.

A clinicopathologic study by Ackermann classified UA into three histologic groups:

- Group I: Luminal UA (tumor confined to the luminal surface of the cyst)
- Group II: Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall)
- Group III: Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium)

Another histologic subgrouping by Philipsen and Reichart includes:

- Subgroup 1: Luminal UA
- Subgroup 1.2: Luminal and intraluminal
- Subgroup 1.2.3: Luminal, intraluminal, and intramural
- Subgroup 1.3: Luminal and intramural

UAs categorized as subgroups 1 and 1.2 can often be managed conservatively through careful enucleation, whereas subgroups 1.2.3 and 1.3, characterized by intramural growths, require more aggressive treatment similar to solid or multicystic ameloblastomas. After enucleation, it's crucial to avoid aggressive bone curettage to prevent deeper implantation of ameloblastoma foci. Chemical cauterization with Carnoy's solution is recommended for subgroups 1 and 1.2.

- Subgroups 1.2.3 and 1.3 have a higher risk of recurrence due to the presence of ameloblastoma tumor cell islands within the cystic wall and potential penetration into surrounding cancellous bone. Late recurrence is common, with an average interval of 7 years. Recurrence rates are also influenced by the histologic subtypes of UA, with those invading the fibrous wall having a higher rate compared to others.

- Recurrence rates also vary based on the type of initial treatment. Lau et al. reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% for marsupialization followed by enucleation (where the lesion reduced in size).

Conflict of Interest

The authors confirm that they have no competing interests.

Consent

The patient provided written informed consent for the publication of this case report and accompanying images.

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