



## CONSERVATIVE MANAGEMENT OF AMELOBLASTOMA IN AN ADOLESCENT PATIENT

### Dental Science

<b>Anoop Kumar Dixit</b>	Post Graduate Student, Carrer Institute Of Dental Sciences And Hospital, Lucknow
<b>Avinash Rathore*</b>	Post Graduate Student, Vs Dental College And Hospital , Bengaluru *Corresponding Author
<b>Gourab Mitra</b>	Post Graduate Student, Vs Dental College And Hospital ,Bengaluru
<b>Arshad Khan</b>	Post Graduate Student, Carrer Institute Of Dental Sciences And Hospital, Lucknow
<b>Rifaquat Ali</b>	Post Graduate Student, carrer Institute Of Dental Sciences And Hospital, Lucknow
<b>Rachana Singh</b>	Post Graduate Student, Saraswati Dentalcollege And Hospital, Lucknow

### ABSTRACT

**Introduction-** Ameloblastoma is an aggressive, benign odontogenic tumour of the jaw with varying clinical features and histologic patterns. The treatment options of en block and segmental resection reduce the chance of recurrence. Resection of the mandible is associated with many complications like loss of jaw bone integrity, deformity, dysfunction, cosmetic and psychological distress. For these reasons, young, growing patients find these treatment options simply unacceptable. An alternative method for young growing patients is the conservative approach.

**Materials and methods-** Enucleation and chemical cauterization of the lesion with carnoy's solution (6% ethanol, 3%chloroform, 1% acetic acid, 1 gm ferric chloride) followed by iodoform dressing routinely.

**Results-** Patient was followed up for 1.5 year. No recurrence was noticed.

**Conclusion-** In young growing patient a more conservative approach with close postoperative follow-up will significantly reduces the morbidity.

### KEYWORDS

Ameloblastoma, carnoys solution, young patient

### INTRODUCTION

Ameloblastoma is described as a benign, slow growing, locally invasive, polymorphic neoplasm that consists of proliferating odontogenic epithelium in fibrous stroma. It originates from the residual epithelium of the enamel organ, with histological appearance is similar to that of the early cap-stage which has not differentiated its later stage of hard tissue formation.<sup>(1)(8)</sup> It was described by Robinson in 1937, as a benign tumour that is "usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent. The World Health Organization (WHO) (1991) defined ameloblastoma as a benign but locally aggressive tumour with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma."<sup>(8)</sup>

It affects most commonly people in China, and Africa.<sup>(1)</sup> The age range of its occurrence is usually between 1<sup>st</sup> and 6<sup>th</sup> decade of life with a mean age of 30.2 years, with male to female ratio of 1:1. It occurs most commonly in the posterior mandible, followed by the maxilla. Despite being so common, it accounts for only about 1% of all tumours involving mandible and maxilla, and 11% of all the odontogenic tumours.<sup>(2)(3)(8)</sup>

Clinically, ameloblastoma is classified into the following groups-unicystic, solid or multicystic, peripheral and malignant, the most common of which is the multicystic variant. Histopathologically, ameloblastoma may be classified as follicular, acanthomatous, granular, basal cell type, desmoplastic, and plexiform types. Desmoplastic type ameloblastoma occurs commonly in the mandibular anterior or premolar region or maxilla. The acanthomatous variant is the least aggressive and a lower recurrence rate, while the most aggressive clinical or pathologic association is seen in the follicular variant. Preoperative diagnostic evaluation includes radiographic imaging (OPG, CT, MRI), and histopathological examination is inevitable. Ameloblastoma originate within the bone, except the peripheral subtype that arise in the gingiva or buccal mucosa.<sup>(1)(8)</sup>

Clinically, it may present as a slow growing, asymptomatic locally invasive tumour, which later can reach size large enough to cause facial asymmetry, displacement of teeth, root resorption, regional paraesthesia depending on the region affected and pathological fractures. Pain is less common but can occur sometimes due to haemorrhage, especially following a traumatic fine needle aspiration, or in case of rapidly growing malignant ameloblastoma.

Radiologically, it may resemble a lytic, scalloped, well circumscribed, slow growing radiolucency, with expansion of cortical plates. It varies from characteristic soap bubble loculations, to unicystic radiolucencies, to subtle appearances such as expanded follicles of erupting teeth. In posterior mandible, it is often seen with unerupted mandibular third molar. It has a typical "soap bubble" or "honeycomb" appearance. The periphery of the lesion is usually smooth. Radiographically, the differential diagnosis of ameloblastoma may be odontogenic keratocyst, aneurysmal bone cyst, fibrosarcoma, or any giant cell tumour.<sup>(5)</sup> MRI is used for ameloblastoma of maxilla mainly, as it helps to visualize its extent to the orbit, paranasal sinuses, and skull base. MRI should be considered in case of desmoplastic ameloblastoma due to their poorly defined soft tissue borders which are often misdiagnosed as a fibro-osseous lesion. In case of malignant ameloblastoma, PET-CT should also be done.<sup>(2)(8)</sup>

Histopathologically, ameloblastoma is characterized by the proliferation of epithelial cells arranged in diverse patterns such as follicular, plexiform, acanthomatous, papilliferous- keratotic, desmoplastic, on a stroma of conjunctive vascular tissue in locally invading structures that resemble the enamel organ at different stages of differentiation. Ameloblastic epithelium may arise from either cells from the rest of enamel organ, or from cells of the sheet of Hertwig's epithelium, or from cell rest of Malassez, or epithelial boundary of an odontogenic cyst, particularly a dentigerous cyst, or from the basal cells of the oral mucosa, or from heterotopic epithelium from other parts of the body, perhaps pituitary.<sup>(6)(8)</sup>

Ameloblastoma has a high recurrence rate (upto 60-80%) if not adequately removed. En-block resection is the treatment of choice with 1-2 cm of bony margin, as the recurrence rate is extremely high. The healthy alveolar mucosa overlying the cortical perforation should also be removed as a margin. On the other hand, resection of mandible has many considerable complications like loss of jaw bone support, facial deformity, dysfunction and psychological distress even after reconstruction.<sup>(3)(9)</sup>

### HISTORY

A 16-year-old female patient reported to the Dept. of Oral and Maxillofacial Surgery, with complaint of swelling in right back region of the lower jaw since 2 years. The swelling was insidious in onset and gradually increased to its present size. There was no history of pain, fever, any discharge from the lesion, and trauma. She gave history of a similar lesion that occurred 3 years back for which she was treated.

Apart from that there was no significant medical history, and habit history of the patient.

**CLINICAL EXAMINATION**

On extraoral examination, facial asymmetry was noted due to the swelling. The swelling was extending from the body of mandible region upto the ramus, anteroposteriorly. Its size was roughly 52 X 33 mm. Intraoral examination revealed, an ovoid swelling on the molar area involving the vestibular, and alveolar region on the mandibular right side. On palpation the swelling was firm, non-tender, non-fluctuant, non-compressible. The surface was smooth, and gingival coloured, stippled, with no surface ulcerations, or pulsations. However, eggshell crackling was present. There were three missing molars (46,47,48).



**Figure. 1- Extraoral photograph of the patient**



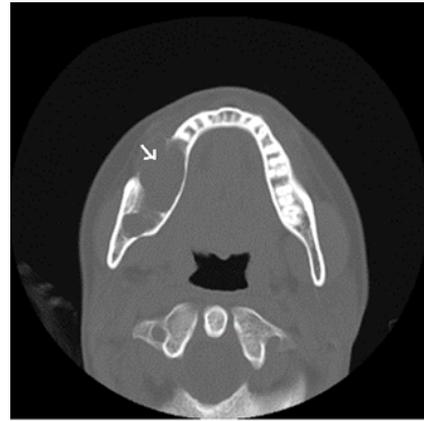
**Figure. 2- Intraoral photograph of the lesion**

**RADIOGRAPHIC EXAMINATION**

On radiological investigation, a well- defined multilocular radiolucency with “soap- bubble” type appearance was seen with well demarcated border, and no matrix calcification. There was no erosion noted in the lingual cortical plate, but there was predominant expansion of the same. The lesion was seen to be extending from the mandibular first molar region, upto the ascending border of the mandibular ramus. However, the lower border of the mandible was intact.



**Figure. 3- Orthopantomogram of the lesion**



**Figure. 4- Axial section of the Computed Tomogram of the lesion**

**INVESTIGATIONS**

Radiographic examination like Orthopantomogram and Computed Tomogram was done, along with routine haematologic examination. There were no significant haematological findings.

**TREATMENT**

After clinico- radiological examination, surgical treatment was planned for this lesion with the provisional diagnosis of ameloblastoma. It is well known that ameloblastoma is an aggressive lesion and the chance of recurrence is high so the treatment options were en-bloc resection or segmental resection. But however, in this case keeping in mind the facial aesthetic concerns of the female patient, enucleation of the lesion was chosen as the treatment plan along with chemical cauterisation. The patient was followed up for a long time afterwards at regular intervals.

After complete haematological investigation, the procedure was carried out under general anaesthesia with an intraoral approach. The lesion was approached through an incision extending from the first molar region to the anterior border of ramus. The lesion was enucleated and Carnoy's solution (6% ethanol, 3% chloroform, 1% acetic acid and 1 g ferric chloride) was applied to the cavity using a cotton pledget for 3 minutes after isolating the surrounding tissues with Vaseline/ glycerine. Following this step, the bony cavity was filled with a betadine- soaked gauze dressing partially protruding from the lesion intraorally for a certain unsutured length. Two days postoperative, the betadine- soaked gauge was replaced with an iodoform impregnated dressing. Iodoform dressing was changed at regular intervals.

The histopathological study confirmed ameloblastoma and revealed its histological variant as Plexiform Ameloblastoma.



**Figure. 5- The lesion curretted out through an intraoral approach**



**Figure. 6- 2nd Day Postoperative**

**OUTCOMES AND FOLLOWUP**

The patient was followed for 1.5 years with an excellent result of uneventful healthy healing with no recurrence. A symmetrical facial profile and satisfactory function was achieved. A good bone growth achieved as compared to the preoperative state.



Figure 7- One year postoperative



Figure 8- Intraoral view (one year postoperative)



Figure 10- Orthopantomogram (8 months postoperative)

**DISCUSSION**

The ameloblastoma is a neoplasm of dental organ that could not undergo differentiation to the point of hard tissue formation due to genetic mutations. It generally presents as a painless, slow growing, locally invasive tumour causing expansion and perforation of both the lingual and the buccal cortical plate with soft tissue infiltration.<sup>(8)</sup>

For ameloblastoma, treatment regimens can be divided into three modalities being conservative that includes enucleation and curettage, marsupialisation and radical surgery which includes en- bloc or segmental resection.<sup>(7)(11)</sup>

Treatment modalities should be determined by the lesion's size, anatomical location, histologic subtype, and anatomical involvement. There is a school of thought, that major segmental or en- bloc resection for ameloblastoma with a minimum of 1-1.5 cm of bony margin and uninvolved gingival surface is the treatment of choice. On the other hand, there is another school of thought advocating a more conservative surgical management by enucleation of the lesion from the surrounding bone.<sup>(12)(11)</sup>

Gardner recommended that the treatment for solid ameloblastoma was to be radical treatment, whereas unicystic ameloblastoma could be

usually cured by curettage. Mandibular resection can prove devastating to masticatory function, deglutition, phonation, oral competence, and facial appearance. Also, postsurgical defects in the maxillary region can lead to hypernasal speech, fluid leakage into the nasal cavity, and impaired masticatory function. Satisfactory reconstruction of complex jaw defects, is therefore a surgical challenge.<sup>(4)(7)</sup>

A retrospective study done in Northern California (Sampson D.E., Pogrel M.A.) in the year of 1999, for specific diagnostic and treatment techniques for ameloblastoma.<sup>(12)</sup>

Anatomical location	Unicystic lesion	Multicystic/solid lesion
Anterior mandible (cuspid-cuspid)	Curettage/enucleation	Marginal resection Small lesion <3 cm, enucleation with peripheral osteotomy
Posterior mandible (bicuspid-condyle)	Curettage/peripheral osteotomy	Marginal resection without continuity defect (1-2.0 cm margin inferior/posterior border) Segmental resection with continuity defect → thinning of inferior/posterior border
Anterior maxilla (cuspid-cuspid)	Partial maxillectomy	Partial maxillectomy
Posterior maxilla (bicuspid pterygoid plate)	Total maxillectomy	Total maxillectomy

Figure 11- Treatment modality based on anatomical position<sup>(12)</sup>

Group I: confined to maxilla without involvement of the orbital floor	Partial maxillectomy
Group II: involving orbital floor but not involving periorbital area	Total maxillectomy
Group III: involving orbital contents	Total maxillectomy + orbital exenteration
Group IV: involving skull base	Total maxillectomy + orbital exenteration + skull base resection

Figure 12- Treatment algorithm for maxillary ameloblastoma<sup>(12)</sup>

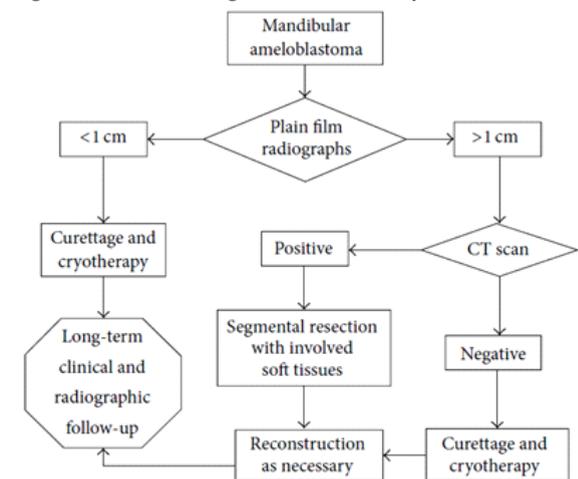


Figure 13- Specific diagnostic and treatment techniques for ameloblastoma<sup>(12)</sup>

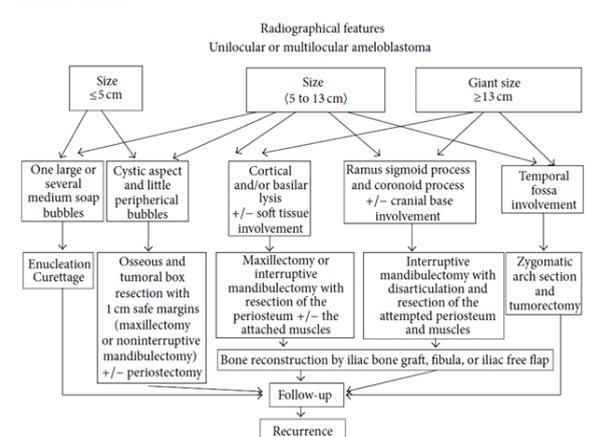


Figure 14- A study done in Pitie Salpeterie Hospital, proposed a therapeutic algorithm for ameloblastoma according to the results and

considering the four main parameters (radiographic presentation, histologic type, size, and location).<sup>(12)</sup>

In this case, the lesion was within 5 cm in greatest anteroposterior dimension, and radiographically it was had a well-defined radiolucency with “soap bubble” appearance, with one large bubble and another medium sized bubble. This according to the retrospective study done in Northern California (Sampson D.E., Pogrel M.A.), pointed towards conservative approach by enucleation and curettage.<sup>(12)</sup> In another study (2015) done by J. Haq, S. Siddiqui et al, demonstrated that the recurrence rate was low by sterilisation of the ameloblastoma cyst cavity with Carnoy's solution.<sup>(9)</sup> In another retrospective study (2017) done by A. Akare, A. Datarkar et al, concluded that enucleation along with the adjuvant therapy has played important role in treating ameloblastoma, the advantage being non-requirement of reconstruction.<sup>(10)</sup> So, it was decided for enucleation of the lesion along with curettage and use of carnoy's solution, to chemically cauterize the region, as that would be the suitable option for this case, keeping in mind the patient's gender, quality of life, concern for facial aesthetics, the type of lesion, and the size and shape of the lesion.

## CONCLUSION

In brief, ameloblastoma is one of the most worrying lesions that Oral and Maxillofacial surgeons may have to deal in their clinical practice. It should be detected and diagnosed as early as possible. Given that this lesion is potentially expansive, destructive, and aggressive, it is essential that proper treatment plan is executed, keeping in mind proper the advantage- disadvantage scale. In case the decision is to treat the lesion conservatively vigorous curettage, peripheral osteotomy, application of Carnoy's solution, or cryosurgery, or resection followed by electrocauterization, should be done in order to further decrease recurrence rates.<sup>(11)</sup>

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