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Review Article

Management of Ameloblastoma -A review

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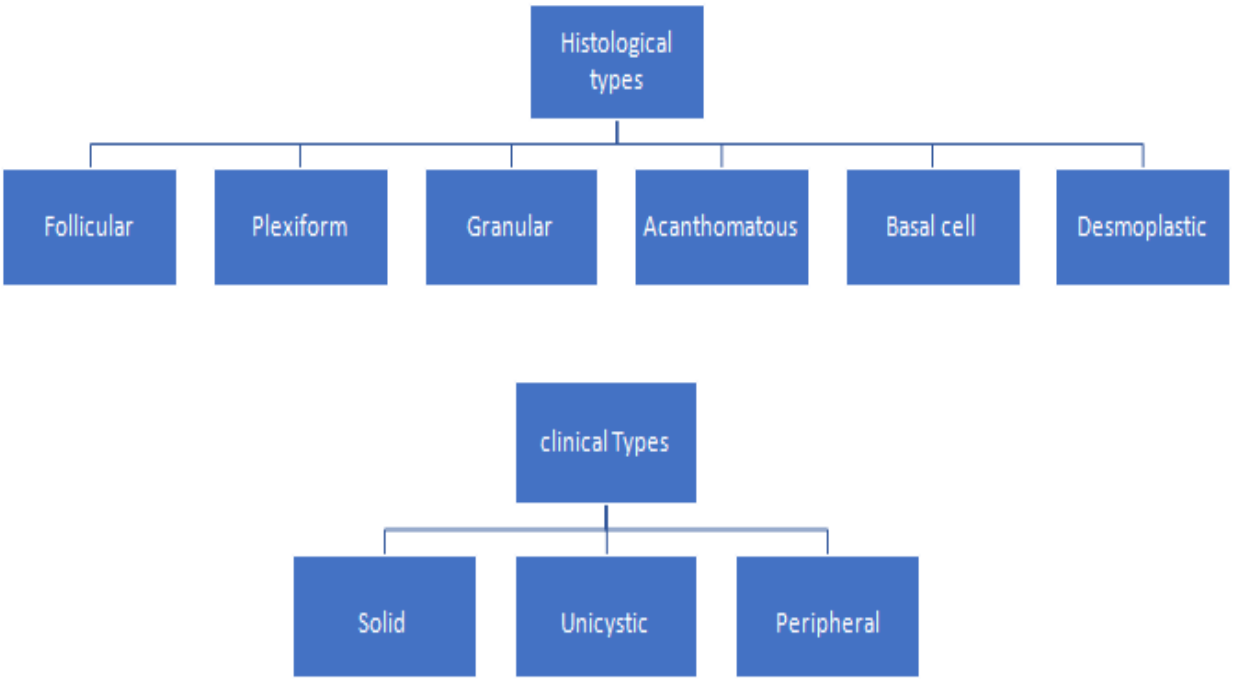
DEFINITION

The ameloblastoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation.

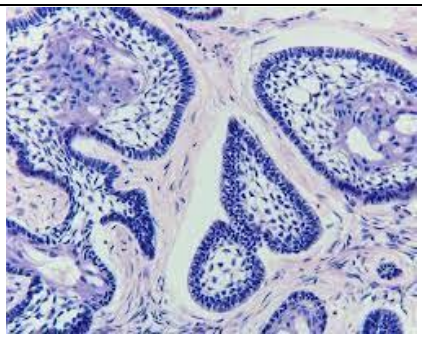
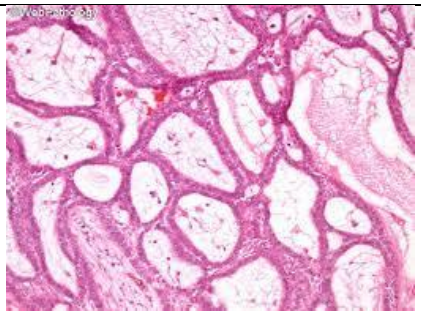
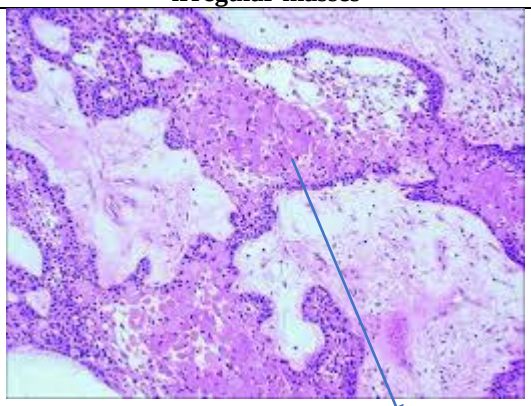
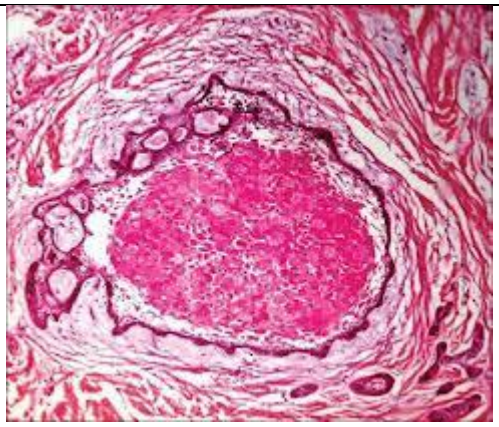
Robinson described the tumour as 'usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent' [1]

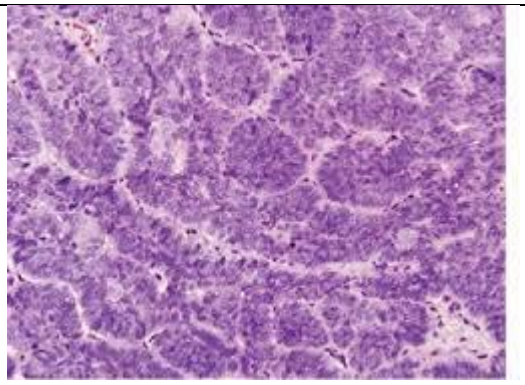
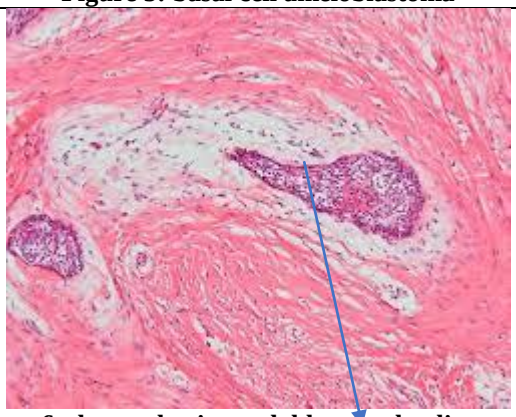
Ameloblastomas are rare, slow-growing tumors of odontogenic origin. Eighty percent of these lesions occur in the mandible.' [2]

TYPES



HISTOLOGICAL TYPES

Follicular	is the most commonly encountered variant, consisting of many small discrete islands of tumour composed of a peripheral layer of cuboidal or columnar cells whose nuclei are generally well polarized[1]	 small discrete islands **Figure 1: follicular**
Plexiform	the ameloblast-like tumour cells are arranged in irregular masses or more frequently as a network of interconnecting strands of cells.[1]	 **Figure 2: plexiform ameloblastoma showing irregular masses**
Acanthomatous	the cells occupying the position of the stellate reticulum undergo squamous metaplasia, sometimes with keratin formation in the central portion[1.]	 **Figure 3: acanthomatous ameloblastoma keratin formation**
Granular cell	a is a rare histopathological entity where there is marked transformation of the cytoplasm of the stellate reticulum-like cells, so that it takes on a very coarse, granular, eosinophilic appearance[1]	 **Figure 4: granular cell ameloblastoma**

Basal cell	bears considerable resemblance to the basal cell carcinoma of the skin[1]	 **Figure 5: basal cell ameloblastoma**
Desmoplastic	a rarely encountered variant, characteristically consists of a dense collageneous stroma that may appear hyalinized and hypocellular[1]	 **Figure 6: desmoplastic ameloblastomahyalinestroma**

CLINICAL TYPES

There are three clinical types of ameloblastoma:

- the solid or multicystic type;
- the unicystic type; and
- the rare peripheral type.

Important considerations in planning treatment include the clinical type as well as the anatomic location of the tumor and the availability of the patient for followup examinations.

The various histologic patterns exhibited by this tumor have no bearing on its biologic behavior[3] The benign nature of these lesions often leads the surgeon to perform simpler extirpative procedures to avoid the potential morbidity associated with larger resections. [2]

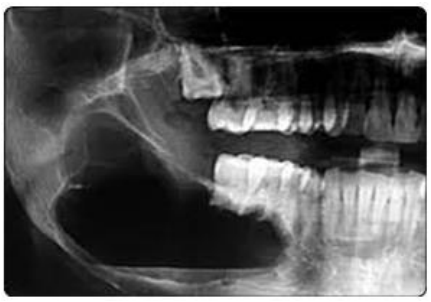
Current treatment options for ameloblastoma include both conservative treatment (enucleation or curettage) and resection. The former is associated with high rates of recurrence, while the latter results in significant facial deformity and morbidity[4]

SOLID

Solid and multicysticameloblastomas have a high recurrence rate following simple enucleation (60–80%). [5]

Mostly this type is diagnosed in young adults, with a median age of 35 years and no gender predilection. About 80% of ameloblastomas occurs in the mandible,frequently in the posterior region.The lesions more often progresses slowly, but are locally invasive and infiltrates through the medullary spaces and erodes cortical bone. If left untreated, they resorb the cortical plate and extend into adjacent tissue[6]

Figure 7: solid ameloblastoma



MANAGEMENT

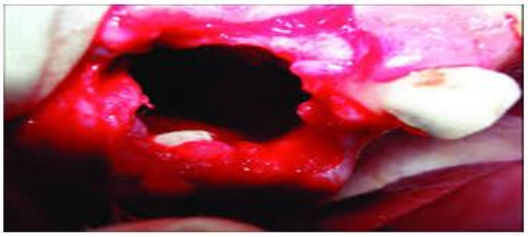
Surgical resection of a solid or multi cysticameloblastoma with 1-cm margins in bone and a margin of one tissue plane in soft tissue is associated with a recurrence rate close to zero. Treatments such as enucleation and peripheral ostectomy or physicochemical treatments including liquid nitrogen or Carnoy's solution have been suggested; initial studies are promising but long-term results are not available[5]

ENUCLEATION

Enucleation with bone curettage was defined as the procedure in which the ameloblastoma was enucleated in conjunction with excision of the overlying mucosa and, subsequently, sufficient bone

curettage. While curetting the bone, 2.5% gentian violet was used on the surface of adjacent healthy bone to ensure removal of the tumor. The colored bone was usually curetted three or four times, for more than 5 mm in depth, by using a large round bur. If an isolated tumor nest was recognized in the cancellous bone during this procedure, additional curettage was performed. When the nerve was exposed in the surgical field, it was lifted out from the bony canal when curetting the bone to avoid damage to the nerve. After the surgical treatment, the patients had clinical and radiographical examinations every year for at least 5 years.[7]

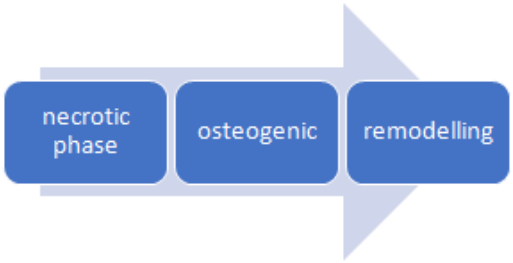
Figure 8: enucleation



CRYOSURGERY

The cryotherapy mechanism consists of eliminating abnormal cells within bone by freezing and devitalizing bone segments after the macroscopic tumor has been removed without the need for radical resection or bone graftinglThe bone segments treated in situ keeps the inorganic matrix, which acts as a perfect autogenous graft.

The effects of cryosurgery on bone tissues were divided into three overlapping stages: a necrotic phase, an osteogenic phase, and a remodeling phase. The initial phase takes place a few days after treatment and results in cell loss and bone tissue necrosis. The osteogenic phase occurs over several weeks during which new subperiosteal woven bone is formed, replacing the necrotic bone. The final phase takes many months and consists of remodeling the formed woven bone and replacing it with vital lamellar bone.



Four basic cryosurgery methods are used in the maxillofacial region

- probe alone,
- probe plus water-soluble jelly,
- liquid nitrogen coil, and
- liquid nitrogen spray.

Infrared thermography has been used to evaluate the freezing bone area of each cryotherapy method[8]

EN BLOC RESECTION OR MARGINAL / SEGMENTAL RESECTION

- If cortical bone is resorbed and penetrated, the resection should include periosteal layer
- A thin inferior border of the mandible in the first procedure may fracture, if a reconstruction plate is not used to span and support the segment. Retention of less than 1 cm in thickness of an inferior border is not practical and second surgical procedure should be opted
- If the complete excision of the tumor is ascertained by clinical and radiographic examination of specimen or intraoperative frozen section, then immediate reconstruction can be undertaken
- If there is uncertainty about resection margins, reconstruction should be delayed until no recurrence is seen, at least after six months postoperatively.
  Adequate soft tissue coverage should be available, if immediate reconstruction is planned
- Immediate reconstruction can be done by using an autogenous free bone grafts (iliac or rib graft) or bank allogenic bone crib and autogenous bone marrow with a reconstruction plate
- Reconstruction plate with or without condylar prosthesis can be used in very old patients, or in cases where secondary reconstruction is planned or where adequate soft tissue coverage is not available
- If sufficient soft tissue is not available locally, a vascularized composite pedicle graft of bone and myocutaneous tissue can be used
- In maxilla—aggressive resection is carried out

Figure 8: Intraoral margin resection in anterior region of mandible

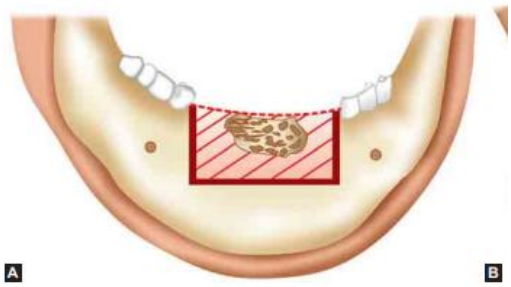
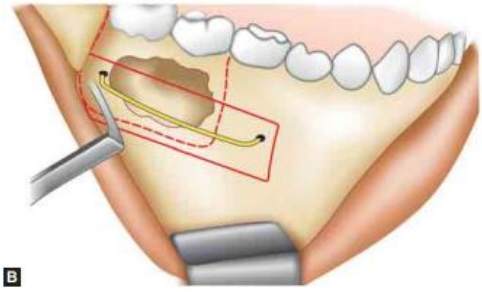


Figure 9: inraotal marginal rresection in posterior region of mandible



Jackson and Callon Forte (1996) have given guide lines depending upon anatomical extents:

Extent	Procedure
Tumor confined to maxilla without orbital floor involvement	partial maxillectomy
Tumors involving the orbital floor, but not the periorbital area	total maxillectomy
Tumor involving orbital contents	total maxillectomy with orbital exenteration
Tumor involving the skull bone—along with skull base resection	along with skull base resection—neurosurgical procedure. Behavior: The multicysticameloblastoma has a recurrence up to 50% during the first 5 years postoperatively. Longterm follow-up is a must

Curettage should never be considered as the treatment modality, since intraosseousmulticystic lesions 'recurrence rate is 55–100% after curettage, and for intraosseousunicystic lesions— 18–25%. The characteristic feature of this tumor is that it microscopically infiltrates bone beyond the tumor-bone interface seen in imaging. A safe margin of uninvolved bone is approximately 2 cm for solid and multicystic lesions. This may mean that important structures like inferior alveolar nerve may be resected en bloc with the tumor

EXTRAORAL APPROACH

It may be necessary for difficult approach. The surgical procedure will be same as described earlier for the marginal resection

1. Segmental resection including the condyle (Disarticulation)
2. The surgical approach is through a combined postramal (Hinds) and submandibular (Risdon) incision placed at least 2 cm below and parallel to the inferior and posterior borders of the mandible

Figure 10: segmental resection including condyle

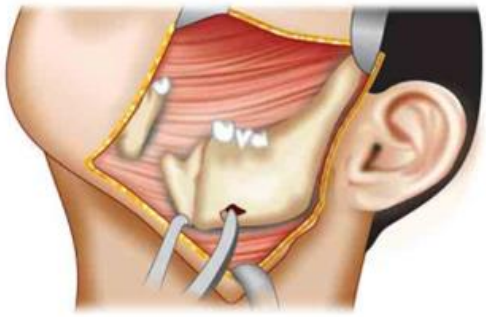
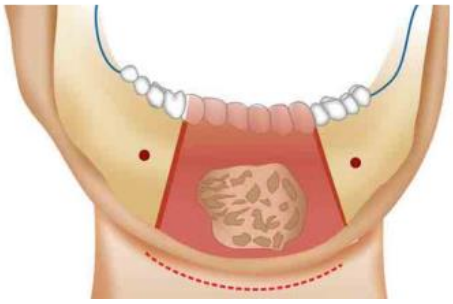


Figure 11: extraoral segmenal resection involving mandibular midline



UNICYSTIC

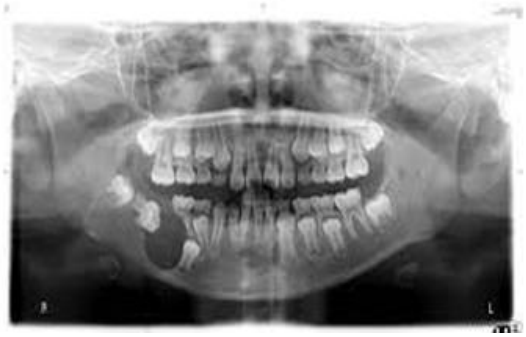
Unicysticameloblastoma usually appears very similar to a non-neoplastic odontogenic cystand is frequently clinically misdiagnosed as dentigerous cyst and odontogenic keratocyst, hence histological confirmation is mandatory.[10]

Unicysticameloblastomas occur most commonly in the second and third decades oflife,

Unicysticameloblastomas are cystic lesions in which one or more of the following features are present:

1) the lining epithelium is ameloblastomatous;
2) one or more nodules of ameloblastoma project into the cystic lumen (the so-called luminal ameloblastoma);
3) ameloblastomatous epithelium of the lining has proliferated into the connective tissue wall of the cyst (one form of the socalled mural ameloblastoma);
4) islands of ameloblastoma are present in the connective tissue wall of an apparently nonneoplastic cyst (another form of mural ameloblastoma)[3]

Figure 12: unicystic ameloblastoma



MANAGEMENT

Various treatment modalities for unicystic ameloblastoma have been used, such as segmental or marginal resection as normally used for conventional ameloblastoma, however, more conservative treatments have frequently been reported.

- ✓ Enucleation and
- ✓ curettage are mostly chosen.

Some clinicians used marsupializationto reduce the size of the lesion, followed by second stage surgery. These treatments can be followed by adjunctive therapy, including cryotherapy, thermal or chemical cauterization and even radiotherapy and

chemotherapy. The reported recurrence rate after treatment of unicysticameloblastoma ranges from 10 to 25% [10]

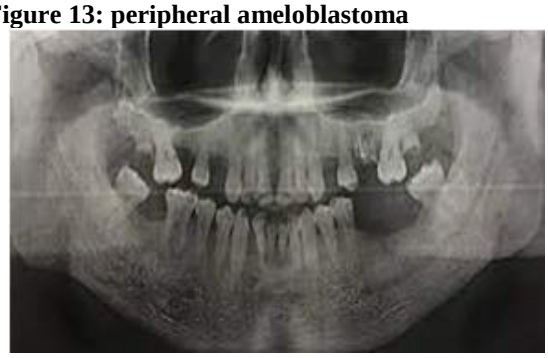
Current studies revealed that the third histologic subtype of unicystic ameloblastoma (a cystic lining showing intraluminal and intramural proliferations) was aggressive and should be treated more radically as a solid ameloblastoma.

Therefore, recommend relatively conservative treatment in initial presentation, such as enucleation and curettage followed by physiochemical treatment or peripheral ostectomy in the mandible and localized resection in maxilla, and reserve more aggressive therapy for any recurrences [11]

Peripheral ameloblastoma

Although histologically identical, this probably represents a different lesion that is in the soft tissue of the gingiva only and is essentially benign. It responds well to local excision.

Figure 13: peripheral ameloblastoma



MANAGEMENT

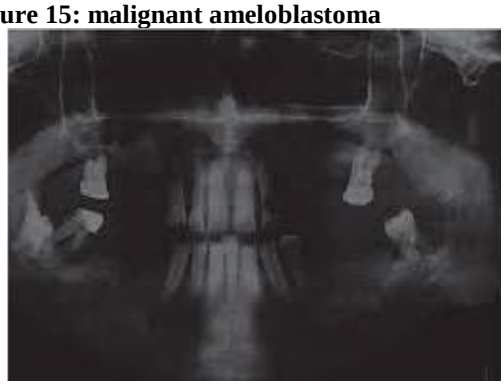
The peripheral ameloblastoma lacks the persistent invasiveness of the solid or multicysticameloblastoma. So far, only one case has been reported as invading bone, but this type of ameloblastoma may cause cupping of the underlying bonc. Theydo not tend to recur if excised adequately, and therefore appropriate treatment consists of *excision with a small amount ofsurrounding tissue, followed by periodic examination of the surgical site*.

This is in fact the usual treatment performed for peripheral ameloblastomasand the histopathologic diagnosis is made only postoperatively. If, on histologic examination, there is any doubt that the lesion has been completely removed, a further excision should be performed through the periosteum. Neither bone nor teeth need generally be removed, but some underlying bone can be excised if the surgeon considers it[3]

MALIGNANT SUBTYPES

The term "malignant ameloblastoma" is applied, when the metastatic tumor resembles primary tumor with no histological transformation. The term "ameloblastic carcinoma" is used, when there has been obvious histological transformation towards malignancy of epithelial component and metastatic tumor resembles to a less well differentiated carcinoma, usually an epidermoid carcinoma. These types are rare, aggressive variants with obvious metastasis and poor prognosis, at times inoperable

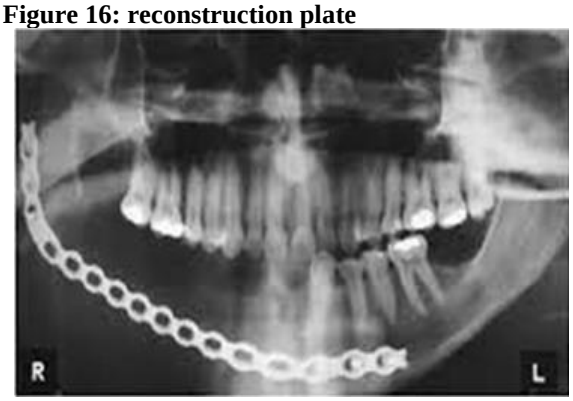
Figure 15: malignant ameloblastoma



MANAGEMENT

Successful treatment is the treatment that renders an acceptable prognosis, causes minimal disfigurement and is based on the behavior and potential of the tumor, the growth patterns of the various physical forms, duration, the anatomic site of occurrence, the clinical extent and size of the tumor and histologic assessment. The treatment modality is also determined considering the age and general health of the patient. Complete eradication of lesion and Reconstruction of the resultant defect [1]

Figure 16: reconstruction plate



AMELOBLASTOMA IN CHILDREN

The tumor is considered a rarity in the young, but the tumor grows slowly and probably starts to develop in childhood[12]

Ameloblastoma is uncommon in the pediatric population, with only 8.7% to 15% of all ameloblastomas in Western countries.

The Asian and African reports show a higher percentage, ranging from 14.6% to 25%.

Because ameloblastoma is uncommon, only a few surgeons have much experience in treating this lesion. The extent of surgical excision of mandibular ameloblastoma has been the subject of debate for many years

Figure 17: ameloblastoma in children

MANAGEMENT

The treatment for ameloblastoma is still controversial and poses some special problems in children. Because of growth of the jaw, the different incidence, behavior, and prognosis of the tumor make the surgical consideration different from adults use the decompression procedure to decrease the extent of lesion and to get optimal specimens for serial section examination. The cystic ameloblastomas were treated conservatively, by decompression, enucleation and peripheral ostectomy, and long-term and regular follow-up. Only when recurring more than twice or according to the patient's wishes, then more aggressive surgeries might be considered[12]

CONCLUSION AND SUMMARY

Ameloblastoma is a rare tumor of odontogenic epithelial origin constituting only 1% of all tumors and cysts of the jaws,[4] but it is the second most common odontogenic tumor.[1] Ameloblastoma affects both genders equally and presents as a painless swelling predominantly in the molar region and ascending ramus. Typically, primary tumor of the mandible appears as a multilocular radiolucency often with buccolingual expansion. Resorption of adjacent teeth and those of unerupted teeth may also be seen. Most cases are reported between 30 and 60 years of age. It is rare before the age of 20 years.[1] Only 1.8% of patients are reported to be 10 years of age[13]

It is essential to obtain an accurate histologic diagnosis from a biopsy before definitive treatment is commenced for a suspected solid or multicysticameloblastoma. On the other hand, unicystic and peripheral ameloblastomas are usually not biopsied preoperatively, because they are generally removed as suspected cysts or epulides.[3]

One approach to treatment of ameloblastoma is often labeled "conservative" and generally consists of curettage or enucleation. The term enudeation should be abandoned in favor of the term curettage. Gardner and Pecakldefined enucleation as the "removal of a lesion by shelling it out intact."[2]

Reconstruction of the defects with bone graft material allows good functional and esthetic outcome and decreases the number of surgeries. For reconstructing the mandible we prefer bone grafts from the iliac crest. The natural curvature and variable bone height offers the possibility of exact reconstruction of the defect.[13]

Postoperative follow-up is very important because more than 50% recurrences occur within 5 years of treatment.[3]Many reports reveal the recurrence time is between 1 and 15 years, and 2–5 years is the most common. For the most accurate recurrence rate findings, all cases should be routinely follow-up a long time because of the insidious biological behavior of ameloblastoma[11]

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