Original Article

The Mandibular Ameloblastoma Saga – An Institutional Experience

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Abstract

Background: Ameloblastoma is a true benign odontogenic neoplasm with many classical histological variants, common being follicular and plexiform types. Being a benign odontogenic tumor of epithelial origin, it is a locally aggressive tumor and has a high potential for malignant transformation as well as metastasis. Ameloblastoma has no established preventive measures although the majority of patients are between ages 10 and 60 years. The treatment of ameloblastoma is focused on surgical resection with a wide margin of normal tissue because of its high propensity for locoregional invasion; but this is often associated with significant patient morbidity. The relatively high recurrence rate of ameloblastoma is influenced by the type of molecular etiological factors, the treatment modality and how early the patient presents for the treatment. **Materials and Methods:** Twenty-five cases reported in our Department of Oral and Maxillofacial Surgery from 2019 to 2021 and were treated with various treatment modalities to minimize post-operative complications and recurrence. **Results:** We evaluated all the cases and found out that it showed a male predilection affecting mainly the posterior region of the mandible. The histologic predilection in our cases showed that follicular type was the most common followed by plexiform. **Conclusion:** Ameloblastoma is the most commonly occurring odontogenic tumor in the mandibular body ramus region in the middle age group (multicystic) and in young age group (unicystic). The best treatment of ameloblastoma is aggressive *en bloc* resection with simultaneous reconstruction.

Keywords: Ameblastoma, Dredging, Enucleation, Recurrence, Resection.

Introduction

Ameloblastoma is a benign epithelial odontogenic tumor that usually exhibits aggressive behavior. It expands cortical bones and has a high recurrence rate. It also causes mobility and displacement of the teeth, as well as root resorption.[1]

Categorized on clinico-radiological features into three types: Solid or multicystic, unicystic, and peripheral.[2]

Various histological variants of ameloblastoma have been described in the literature such as follicular (most common), plexiform, acanthomatous, granular (most aggressive), basal cell, and desmoplastic among the solid type and unicystic variants include mural, intraluminal, and luminal.[3,4] Reports of variants such as clear cell, papilleferous, keratoameloblastoma, and hemangiomatous ameloblastoma are also there. Desmoplastic and hemangiomatous ameloblastoma are exceptions with the former thought to be more aggressive and the later due to its extreme vascular component carries surgical complications.[4,5]

Aims and objectives

Purpose of study

The purpose of the study is to retrospectively analyze present a series of 25 cases of mandibular ameloblastoma treated in the Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research emphasizing on the various treatment modalities of ameloblastoma and the different clinical and histopathological types.

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Materials and Methods

We retrospectively evaluated 25 patients of ameloblastoma of mandible.

Time period

The study period was from July 2016 to January 2021.

Study place

The study was conducted at the Department of Oral and Maxillofacial Surgery, Haldia Institute of Dental Sciences and Research.

Inclusion criteria

We included patients who were: (a) Incisional biopsy proven ameloblastoma and (b) ASA I and ASA II class of patients.

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Exclusion criteria

(a) ASA III and ASA IV patients and (b) those patients ready to come for follow-up were excluded from the study.

All the patients reported with slow growing painless swelling on the left or right region of the mandible involving either body, or ramus, or angle and sometimes all the three regions crossing the midline.

We have included some images of the cases which were done in our department (Figures 1-3).

Procedure

The treatment regimens in our cases were divided into three modalities (a) conservative that includes enucleation and dredging, (b) marsupialization, and (c) radical surgery which includes resection with or without continuity defect.[6] For all the cases of multicystic ameloblastoma, radical surgery was done. Resection of the jaw approximately 1.5–2 cm beyond the radiological limit was done, to ensure that all the "microcysts" and "daughter cysts" are removed.[7] Because of the large size of these lesions, mostly of whom crossed the midline, we have used free fibula flap for reconstruction of the defect in 12 of our cases. For the unicystic variant, we performed enucleation and dredging.[8]

All the cases were followed up for minimum of 24 months post-operative.

Statistical analysis

We performed student *t*-test to statistically evaluate the recurrence rate.

Results

We evaluated all the cases and found out that it showed a male predilection affecting mainly the posterior region of the mandible. Multicystic ameloblastomas affected patients between the third and fourth decades of life whereas the unicystic ameloblastoma affected more young patients, generally in the second decade of life. The histologic predilection in our cases showed that follicular type was the most common followed by plexiform. The luminal type of unicystic ameloblastoma was also seen in four of our cases, followed by one mural and one intraluminal case.

All our patients those who had shown solid variety of ameloblastoma were treated with resection followed by reconstruction. Furthermore, in the case of the mural variety of unicystic ameloblastoma, we had done segmental resection. For all other varieties of the unicystic type, we performed enucleation followed by dredging.

Our patients are also under follow-up with no evidence of recurrence except for one case where we experienced a complication of reconstruction plate loosening due to infection on the operated site, which we resolved it successfully. Furthermore, in one more case, the lesion showed signs of recurrence after 1 year following, we performed a further incisional biopsy that revealed granular ameloblastoma and we performed marginal mandibulectomy followed by reconstruction with a reconstruction plate.

We performed student *t*-test to statistically evaluate the recurrence and found out that (P=0.12) which was statistically insignificant.

Discussion

Clinically, an ameloblastoma is an odontogenic tumor that can cause local bone destruction and can be metastatic. They

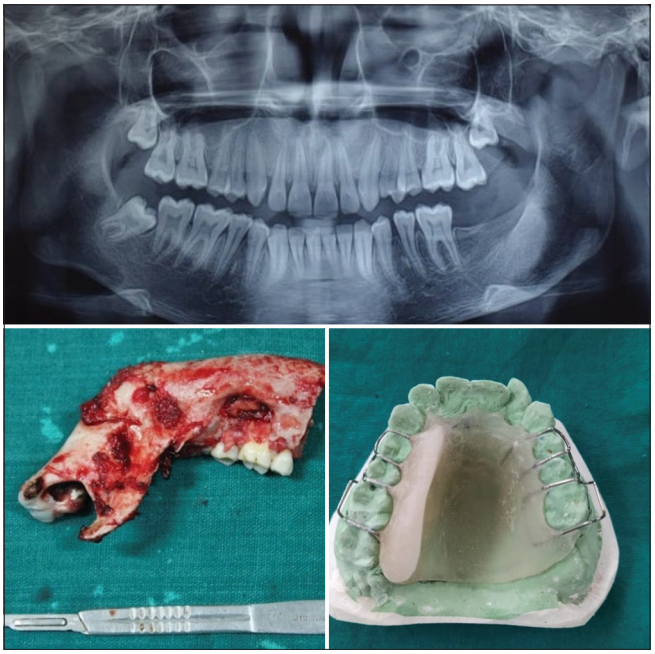

Figure 1: Hemimandibulectomy and reconstruction with titanium miniplate done on the left mandible and post-operative guiding flage given

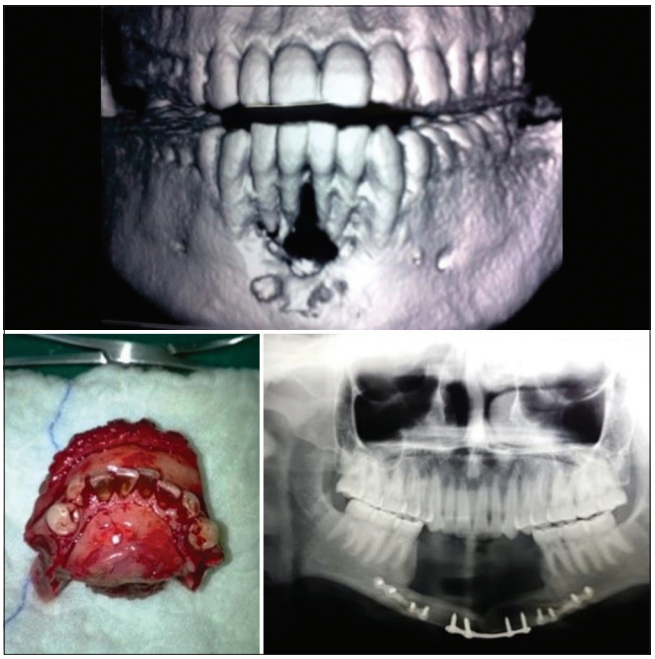

Figure 2: Wide local excision of lesion and reconstruction with free fibular graft

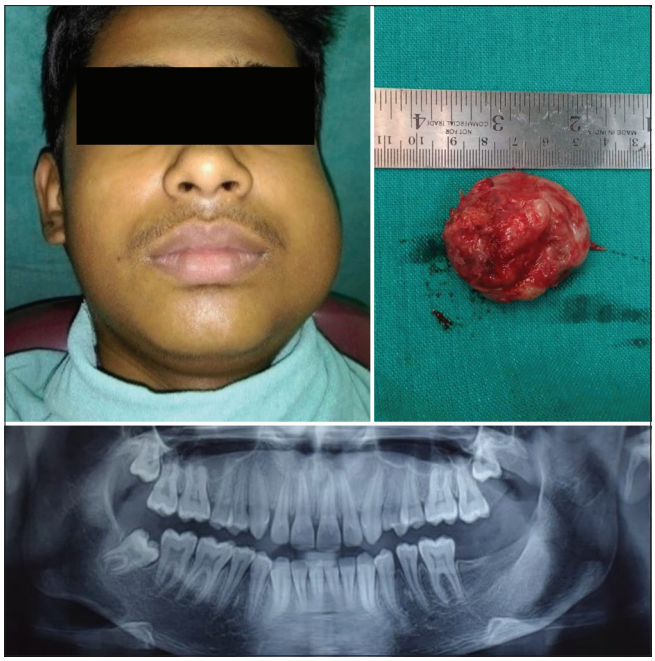

Figure 3: Enucleation of lesion at the left side of mandible

are slow growing and progressive. They are painless, difficult for the patient to perceive in the initial stages, and, while they develop, cortical resorption occurs because of the compression produced by growth, making them palpable; in most cases, diagnosis is complemented by radiography.[9]

The solid or multicystic form is far aggressive than its unicystic counterpart and its capacity to infiltrate the bone trabeculae and requires a more radical treatment than the unicystic type, with a relatively higher rate of recurrence.[10]

The radiographic aspect is a multilocular radiolucent lesion and described as "soap bubble" or "honeycomb."[11]

Radiographically, the unicystic variant has a unilocular radiolucent mass, which in most cases surround the crown of a tooth that has not yet erupted and is commonly mistaken for a dentigerous cyst. Peripheral ameloblastoma is one more type which is found in 1% cases affecting posterior alveolar and gingival mucosa. This lesion has a good prognosis if it is removed at an early stage when it is easily detected clinically, and, because the cortical bone is still preserved, it is a barrier to bone invasion by the peripheral ameloblastoma.[10]

There are different methods of mandibular reconstruction of large defect with microvascular reconstruction using donor site from fibula, iliac crest, and scapula.[12]

In our study, male predilection is more (60%) compared to female (40%) which supports the study of Small *et al*. (1955),[13] whose result showed the ratio – M: F to be 52:48. Furthermore, in accordance with and Vohra *et al*., our study also showed that posterior region of mandible is most commonly involved.[7]

Here, in our cases of mandibular ameloblastoma, we opted for the different treatment modalities surgically performed which depended on the type and pattern of the ameloblastoma as we know that they show various biological behaviors, ranging from cystic expansion to more aggressive infiltration of adjacent tissues. The architectural pattern of ameloblastoma is such that the border of tumor within cancellous bone lies beyond the apparent macroscopic surface and radiographic boundaries of the lesion.[14]

Sammartino *et al*. advocated conservative approach in unicystic types because of low morbidity associated with these procedures.[8] Many authors have recommended enucleation with preservation of periosteum which is important for bone regeneration especially in children.[15]

Reconstruction with free fibula flap offers lot of advantages in mandibular reconstruction. It allows for transfer of bone, soft tissue, and skin in a one-stage procedure using only one donor site, fibula flap allows for a skin paddle up to 25 cm in length and 5 cm in width, and bone up to 25 cm. Furthermore, double barrel fibula offers an option of placing dental implant supported prosthetic rehabilitation in a later stage. Blood supply to fibula is both intraosseous and segmental; therefore, multiple osteotomies can be made. Postoperatively very good esthetic and functional results were obtained with this type of reconstruction. Hence, reconstruction with free fibula was our most preferred method.[16]

We preferred reconstruction plate in cases of patients having comorbidities such as diabetes and poor fibular vascular supply through color Doppler preoperatively.

Outcome and recurrence

The chance of recurrence seems to be more dependent on the method of surgical treatment. In general, though annual follow-up for at least 10 years is recommended. Few authors have recommended annual follow-up for 5 years and thereafter once in every 2 years till 25 years.[6]

Short-comings of the study

i. Long follow-up recommended. We could only pursue 2 years
ii. More larger sample size is recommended for further evaluation.

Conclusion

Ameloblastoma is the most commonly occurring odontogenic tumor in the mandibular body ramus region in the middle age group (multicystic) and in young age group (unicystic), yet, other differential diagnosis such as odontogenic keratocyst, odontogenic myxoma, and central giant cell granuloma has to be ruled out with the help of advanced radiodiagnostic tools and histopathological examination. The best treatment of ameloblastoma is aggressive *en bloc* resection with simultaneous reconstruction using various options of the treatment modalities depending on the age group and aggressiveness of the lesion. Role of enucleation and dredging is also successful in unicystic luminal and intraluminal variant and in younger age group.

Declarations

Funding

None.

Conflict of interest

None declared.

Ethical clearance no - HIDSAR/PG/2021/A718.

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