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Controversies in management of Ameloblastoma

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Abstract
Introduction
Ameloblastoma has its inclusion in a group of odontogenic tumours that originate from the odontogenic remnants of various tissues which remain trapped in the bone or soft tissue. Management of Ameloblastoma occupies a space in our specialty where the treatment protocol still remains controversial. Various authors have debated over the best approach to treat this tumour. Considering the benign nature of the tumour, it is advisable to offer different treatment options to the patient. The decision whether to go for a conservative or radical approach should also be based on various clinical entities like age, aesthetics, aggressiveness of the tumour etc.
Materials and method
This is a retrospective study of 9 cases of ameloblastoma diagnosed and treated with Enucleation, peripheral ostectomy and chemical cauterization in our institution. Both males and females with a mean age of 24 years were included in the study. All the patients were followed up for a minimum of 6 months and orthopantomograms were used to compare the bone formation pre and post surgery.
Result
All patients treated in this case series reported for review with good bone formation except for one in whom recurrence was noted in themandibular angle region.Another patient in whom a reconstruction plate was placedreported with a draining sinus tract, the radiograph revealed good bone formation and the patient was treated with implant removal and sinus tract excision.No significant treatment related complications were noted.
Conclusion
Conservative treatment if done adequately without leaving any traces of diseased tissue particularly in young patients will reduce morbidity and will not interrupt growth and function thereby improving the quality of life.
Key words: *Resection, Enucleation, Cauterization, Peripheral ostectomy.*

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I. Introduction

Ameloblastoma is a benign, slow growing, locally aggressive tumour originating from the tissues or remnants of odontogenic epithelium. In the maxillofacial region mandible is the most commonly involved structure. Ameloblastoma usually occurs in the middle age group with highest incidence noted in the 3rd decade of life.

Ameloblastoma still remains one of the controversialtumours in terms of treatment due to its distinct aggressive biological behavior despite umpteen number of studies. Different treatment modalities have been offered by different surgical disciplines and different researchers which include aggressive managements like segmental or enbloc resection with 1 cm to 1.5 cm marginal clearance clinically and radiographically, conservative management which includes marsupialization, cryotherapy, enucleation and excision with peripheral ostectomy. [1]

II. Materials and method

A total of 9 patients who were confirmed post biopsy with Ameloblastoma were treated in the Department of oral and maxillofacial surgery during the year 2016 and 2017.Both males and females with a mean age of 24 years were included in the study. Patient falling under ASA I, II and III with one or both the cortices intact were included in the study. Pregnant women and grossly destructed bone were excluded from the study. All the patients were treated with enucleation, peripheral ostectomy and chemical cauterization. Pre-operative post-operative orthopantomogram was taken to assess the bone formation after treatment. All the patients were followed up for a minimum of 6 months.

III. Surgical protocol

The surgical protocol in our study includes enucleation of the tumour mass, peripheral ostectomy using round or vulcanite burs followed by chemical cauterization with Carnoy's solution. In patients with intact but thinned out bone cortices following the procedure,reinforcement with a mini or a reconstruction plate was done to prevent any pathological fracture.An intraoral approach was employed for all the patients except for one where an extraoral approach was used to enucleate the tumour mass and the bone was reinforced usingreconstruction plate. The areas involved were the mandibular ramus, body, parasymphysis and symphysis regions. Primary closure of the surgical site was done in all the patients. The procedure was executed in select patients with predefined inclusion criteria. The patients were followed for a minimum of 6 months.

IV. Results:

Out of the 9 patients operated, 7 patients showed evidence of adequate bone formation with no signs of recurrence in the operated site at the end of 6 months(Fig. 1-4). No intraoral opening or breach was seen in any of the cases. There was no evidence of any extraoral swelling. In one patient who required extra-oral approach and reconstruction using reconstruction plate reported back with an extra-oral draining sinus tract, the radiograph showed evidence of resorption around the screws with good bone formation in the enucleated site. The patient was taken up for surgery for excision of the sinus tract and removal of reconstruction plate. One patient reported back after 8 months for review, the radiograph revealed a recurrence in the angle region. The recurrent lesion was smaller than the primary lesion and was treated with the same surgical protocol (Table 1). All the patients were advised to report for review every 6 months for a minimum of 5 years.

Table 1. List of patients operated with conservative approach.

S. No	Age	Sex	Area of involvement	Approach	Recurrence
1.	13yrs	Male	Left ramus and body	Intraoral	No
2.	15yrs	Female	Right ramus and body	Intraoral	No
3.	27yrs	Male	Right body	Intraoral	No
4.	19yrs	Male	Left body	Extraoral	No
5.	25yrs	Male	Right body and parasymphysis	Intraoral	No
6.	37yrs	Female	Symphysis and left parasymphysis	Intraoral	No
7.	16yrs	Male	Right ramus and body	intraoral	Yes (Angle)
8.	47 yrs	Male	Right body of mandible	intraoral	No
9.	7 yrs	Male	Right body of mandible	intraoral	No

Pre and post operative radiographs

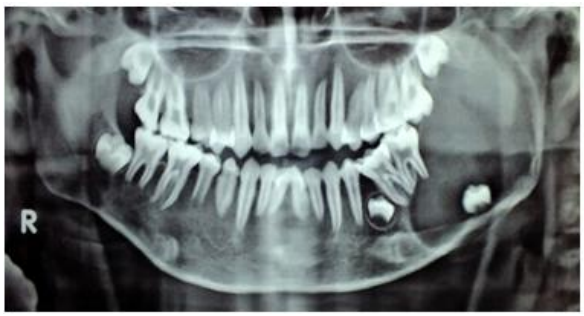

Fig. 1 showing the tumour involving the left ramus and body of the mandible.

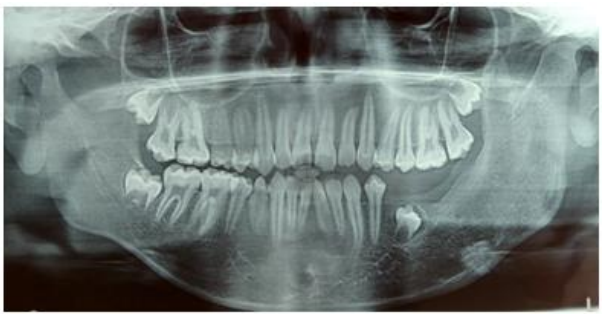

Fig.2 showing the 6 month postoperative radiograph with good bone formation withoutany signs of recurrence.

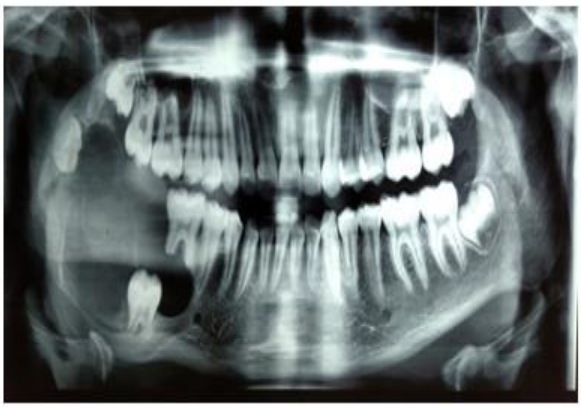

Fig. 3 Showing the tumour involving the right ramus and body of the mandible.

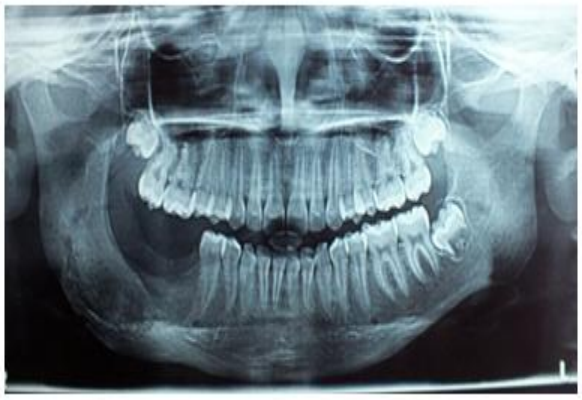

Fig.4 showing evidence of good bone formation 6 months postoperatively without any signs of recurrence.

V. Discussion

Introduced as Adamantinoma by Malassez in 1885, Ameloblastoma, a benign tumour is reported to constitute about 1-3 % of tumours of the jaws.[2]The term Ameloblastoma was given by Ivy and Churchill in 1930.[3] Ameloblastoma is an invasive tumour with a malignant potential. Despite umpteen numbers of studies, the tumour still remains controversial in terms of its treatment. Multicenter collaborative investigations are required to evaluate treatment approaches to ameloblastoma objectively. Until therapeutic guidelines are developed objectively,it remains a clinician's responsibility to formulate a surgical treatment plan that is individualized and patient centered and not based on an arbitrary surgical algorithm.[4]

Ameloblastoma was considered to be a radio-resistant tumour earlier but many authors advocate radiotherapy. Robinson reported one of the first series, in which 18 patients were treated with radiotherapy alone, 13 patients (72%) developed a local recurrence.[5]Sehdev et al reported a series of 11 patients treated with radiotherapy. Initial response was seen in some patients, eventually a local recurrence was seen in most of the patients.[6]Gardner reported on 3 patients treated with radiotherapy, all 3 responded initially but later recurred.[7]More studies are needed to better understand the effectiveness of radiotherapy. Reports on chemotherapy with platinum based agents, cyclophosphamide and methotrexate being tried for the treatment of Ameloblastoma are also available. Ramadas obtained partial response after 13 cycles of combination chemotherapy associating Cisplatin and Cyclophosphamide administered for lung metastasis.[8] Lanham stated that chemotherapy did not show any results including doxorubicin, methotrexate etc, but more studies are needed with regards to chemotherapy.[9]

Surgery is the standard treatment for ameloblastoma. Historically, the extent of resection has been controversial, comprising of two surgical options: ''conservative'' vs. ''radical''.[10]The former involves enucleation/chemical cauterization of the bony cavity, while the latter involves a radical approach that may be segmental or en bloc resection with appropriate margins. The selection and success of surgical options depend on the careful patient's evaluation, accurate history, radiographs & special imaging (CT), good pre-surgery histopathological reporting.

Radical surgery being the current trend for the treatment of ameloblastoma is followed by immediate bone reconstruction to improve quality of life. Proponents of radical approach are of opinion that although these tumours are histologically benign, they are locally aggressive.[11]Chidzonga stated that recommended treatment for ameloblastoma in children should be radical resection 0.5 to 1 cm more than what appears to be normal bone.[12]Arotiba et al also employed radical resection to be the method of choice.[13]The authors suggested that for solid-multicystic ameloblastoma of the mandible, resection of the jaw should be approximately 1.5-2 cm beyond the radiological limit to ensure that all the microcysts and daughter cysts are removed.[9]

In a study conducted by Muhammad UsmanAkhtar et al, 52 patients of ameloblastoma were operated. 38 cases were managed conservatively with marsupialization followed by enucleation (Group A' 15 Patients) and enucleation with peripheral ostectomy (Group B' 23 Patients). Group C' that is 14 Patients were treated aggressively by resection. In conservative treatment regimens Carnoy's solution was applied after enucleation of the tumour whereas, the patients of aggressive surgery were operated with minimum 5mm safety marginal clearance of the tumour. The recurrence rate with average four years follow up was 0.0% for resection, 13.33% for marsupialization followed by enucleation and 8.69% for enucleation with peripheral ostectomy. The results

were encouraging for unicystic ameloblastoma treated patients (Group A' & B'), in best interest of jaw bone contour preservation.[14]

In a systemic review conducted by Lau et al,they found that resection results in the lowest recurrence rates when adequate bone margins are removed.Despite the high success rates of resection, conservative treatment in order to optimize quality of life is generally favoured. The morbidity associated with a radical approach is associated with serious cosmetic, functional, aesthetic problems and donor site morbidity.They also found that enucleation alone yielded the highest recurrence rate among all treatment modalities, the reasons being that the lining of the tumour is inadequately removed especially in posterior maxilla and chances of tumour remnants being left out in complex anatomical structures.[15]

Current opinion regarding treatment of ameloblastomas is essentially based on case reports, anecdotal evidence, retrospective reviews, and histological evidence. There are not many largescale studies with long-term follow-up results. Sammartino et al suggested conservative treatment of large ameloblastoma as it caused 'low morbidity'. According to the them, radical treatment is associated with seriouscosmetic, functional and reconstructive problems.[16]

It has been reported that the recurrence of an ameloblastoma in large part reflects the inadequacy or failure of the primary. As stressed by most of the studies, inadequate removal of the tumour will result in recurrence. Recommended treatment for recurrence is radical surgery, particularly with maxillary ameloblastoma.[17]

In this study, 8 patients reported back with adequate bone healing, 7 out of which had no signs of any recurrence, mucosal openings or infections. One patient in whom an extraoral approach was used and reconstruction plate was used to prevent pathological fracture as the remnant bone had thinned out. This patient reported with a draining sinus tract in the mandibular angle region, however there was no problem seen with the bone healing, adequate bone had formed and the removal of reconstruction plate and excision of the sinus tract proved helpful. Out of the 9 patients, one patient recurrence was noted. The size of the recurrent lesion was small when compared to the primary lesion and was treated again with the same surgical protocol.

As stated by Olaitan et al, the recurrence of ameloblastoma reflects the inadequacy or failure of the primary surgical procedure. Some patients reported with a recurrence in the maxilla who had a primary in the mandible, this unusual appearance in the maxilla has been either stated to be denovo, independent and without any relation to the primary tumour or a result of implantation of tumour cells into mucoperiostel flaps sometimes raised for massive lesions of the ascending ramus.[18,19] Another reason stated is insufficient inclusion of apparently normal bone or because of some of the tumour remained in the less accessible regions.[18]

Most of the authors recommend a minimum follow up period of 5 years after the treatment. Recurrences can occur up to 21 years after treatment however most of the recurrences have been reported within the first five years after treatment.[15]

Implantation of tumour cells into the adjacent soft tissue and difficulty of removal of the tumour from the anatomically complex structures. Blind resections when carried out in inaccessible regions can also lead to recurrence. In order to discover recurrences in time, postoperative examination of the patients is a must and should be done in short intervals of time.Haq J et al in their study reported a case of unicystic ameloblastoma operated with Enucleation and chemical cauterization with no dental extraction's.[20] The patient reported back with a recurrene 3 years later and was treated with the same procedure with extraction of the involved teeth. No recurrence been noted in the same till date, hinting towards the compulsory extraction of the involved teeth owing to the difficulty in removing the tumour from complex interradicular and interdental regions.[20]

VI. Conclusion

Most studies showed that the prognosis for ameloblastoma is more dependent on the method of surgical treatment. Resection with some safe margin (marginal, segmental or composite resection depending on the site and size of the lesion) is the most preferred method for treating ameloblastomas to prevent recurrence. However the morbidity associated with a radical approach is associated with serious cosmetic, functional, aesthetic problems and donor site morbidity.Conservative treatment if done adequately without leaving any traces of diseased tissue particularly in young patients will reduce morbidity and will not interrupt growth and functionthereby improving the quality of life.Further long term studies with bigger sample size are required with regards to conservative management.

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