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ORIGINAL ARTICLE

Ameloblastoma Management: "Horses for Courses" Protocol

Suresh Menon[1] · Veerendra Kumar[1] · S. Archana[1] · Priyangana Nath[1] · Satyapriya Shivakotee[1] · Mahbubul Hoda[1]

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Abstract
Purpose The purpose of the article is to review 45 cases of ameloblastoma treated in a tertiary care centre depending on the extent of the pathology, in terms of recurrence and morbidity of the patients.
Materials and method This was a retrospective study of patients who underwent treatment for ameloblastoma between 2009 and 2018 at Vydehi Institute of Dental Sciences, Bangalore. During the first phase of 4 years, the focus of the treatment was on avoiding any recurrence, and therefore, resection followed by reconstruction with reconstruction plates was the only modality used in ten patients. However, from 2014, it was decided to treat each patient based on the extent of the lesion and decide on either conservative management in the form of enucleation followed by peripheral ostectomy and chemical cauterisation or resection with safe margins and reconstruction with reconstruction plates.
Results The study sample consisted of 45 patients, and the ages ranged from 12 to 65 years with an average of 36. There were 30 males and 15 females. In the first phase of treatment protocol adopted, ten patients underwent resection. In the later period, 18 patients were treated by conservative methods and 16 patients were treated by radical management. There were only three recurrences over a period of 3-year follow-up in the group treated conservatively.
Conclusion Considering the benign nature of the tumour and the morbidity after resection, patients, most of whom are in the younger age group, can still be subjected to conservative treatment provided they are followed up for a long period thus assuring them of a better quality of life.

Keywords Enucleation · Carnoy's solution · Recurrence · Ameloblastoma · Resection

Introduction

Ameloblastoma is a common benign aggressive odontogenic tumour arising from the dental epithelium. They could arise from the dental lamina or basal cells of the oral epithelium, or from cells that differentiate to mimic the ameloblast. They account for 1% of tumours [1] that affect the oral–maxillofacial complex and 13–58% of odontogenic tumours [2]. Approximately, 85% of ameloblastomas arise in the mandible, particularly in the molar and ramus areas [3].

They are classified into four types based on clinical, histological and radiographic findings: unicystic, multicystic, peripheral and desmoplastic. Each type has a distinct clinical behaviour and requires different forms of treatment. The diagnosis is generally made through a

✉ Suresh Menon
psurmenon@gmail.com

Veerendra Kumar
drveeru07@gmail.com

S. Archana
drarchana.dr.s@gmail.com

Priyangana Nath
priyangana17@gmail.com

Satyapriya Shivakotee
drsatyapriyashivakotee@gmail.com

Mahbubul Hoda
mahboobhoda@gmail.com

[1] Department of Oral and Maxillofacial Surgery, Vydehi Institute of Dental Sciences, Bangalore 560043, India

combination of imaging examinations and biopsy to determine the histological type.

Treatment is classified as conservative or radical. Conservative treatment includes enucleation, curettage and surgical excision with peripheral ostectomy or other adjuvant therapy, such as cryotherapy or use of Carnoy's solution. Radical treatment consists of bone resection. There are unique characteristics of this tumour. They are slow-growing tumours with an affinity for bone, not soft tissue, and they have a high rate of recurrence (up to 60–80%) if not adequately removed [4].

Materials and Method

Due to overpowering literature on high rates of recurrence of the tumour, a standard complete removal strategy of resection was employed from 2009 to 2013 ensuring a minimal chance of recurrence. This, however, resulted in many patients ending up with compromised function due to loss of large segments of mandible, replaced with only a reconstruction plate that provided only form to the area. Most of the patients were either reluctant to undergo extensive bony reconstruction with microvascular bone grafts or could not afford the treatment. A total of ten patients underwent this procedure.

From 2014, in view of the morbidity associated with this modality, it was decided to treat the cases based on the extent of the pathology. All cases with extensive involvement of the mandible including complete loss of bone with perforation and soft tissue involvement were subjected to resection with safe margins. Cases that had the lesion completely intraosseous with bony continuity present were subjected to a treatment of enucleation, peripheral ostectomy and chemical cauterisation with Carnoy's solution. In some cases where there was a probability of pathological fracture in future, reinforcement with a reconstruction plate was also done in these cases. All cases were primarily closed after the procedure.

Results

There were 45 patients treated for ameloblastoma during this 9-year period. The age of patients ranged from 12 to 65 with 3 patients (6.66%) in the age group 10–20, 25 patients (55.5%) in the age group 21–35 and 17 patients (37.7%) in the age group above 35 years (Fig. 1). The average age of the patients was 36 years. Twenty-five of the cases (55.5%) involved the angle/body/ramus region, 10 (22.2%) were seen in the body region, 9 (20%) in the anterior mandible and 1 (2.22%) in the ramus (Fig. 2).

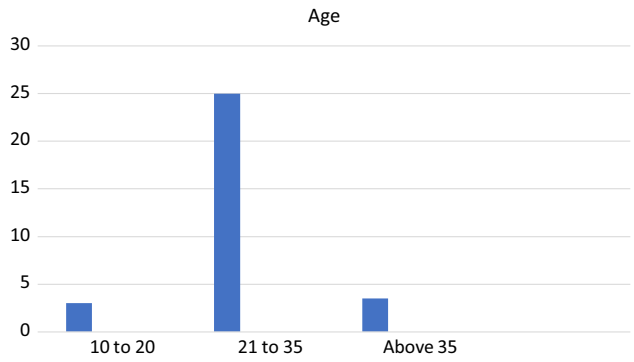


Fig. 1 Age of the patients

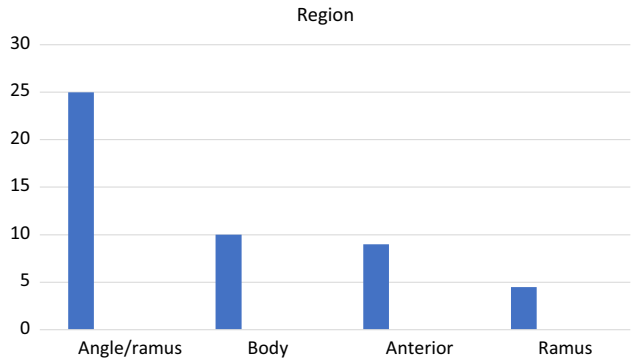


Fig. 2 Region of involvement

In the period from 2009 to 2013, 10 cases (22.2%) were subjected to resection irrespective of the extent of the pathology. A retrospective review of the preoperative radiographic images revealed that 5 of the patients had good bone involving the inferior/posterior border of the mandible and they could have benefited from a more conservative approach. Two of these patients had reconstruction with fibula and one with iliac graft.

From 2014 when it was decided to customise the treatment based on the extent of the pathology, 35 patients (77.7%) were treated. Of these, 16 patients who had extensive involvement of the bone with large-scale perforation or were diagnosed as ameloblastic carcinoma were subjected to resection with safe margins. Two of these underwent reconstruction with fibula and one with iliac graft.

In 19 patients (42.2%), a conservative approach was employed (Picture 1). The protocol included access through either intra-oral or extra-oral incisions, enucleation of the tumour, peripheral ostectomy with acrylic burs, chemical cauterisation with Carnoy's solution and primary closure. When the inferior alveolar neurovascular bundle was exposed, it was covered with vaseline during application of Carnoy's solution and the period of exposure

restricted to 3 min. Only in one case was an additional reinforcement done with reconstruction plate.

Picture 1 .

All teeth that were found to be involved within the pathology were extracted (Picture 2). The cases were followed up for a period ranging from 2 to 4 years. In the 19 cases that were treated conservatively, 4 were lost to review. The other 15 cases showed excellent bone remodelling and filling up of the defects. In 3 cases, there was evidence of recurrence, seen as radiolucent lesions of sizes 2–3 cm within the reparative bone in the radiographs (Picture 3). These were treated by local ostectomy and chemical cauterisation.

Histologically, 14 cases were plexiform type, 7 follicular, 6 acanthomatous, 6 unicystic, 6 desmoplastic and 6 ameloblastic carcinoma (Fig. 3). All 6 of the latter were subjected to resection.

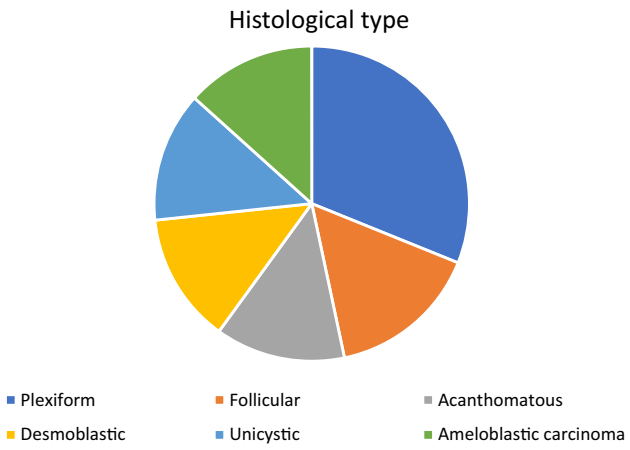
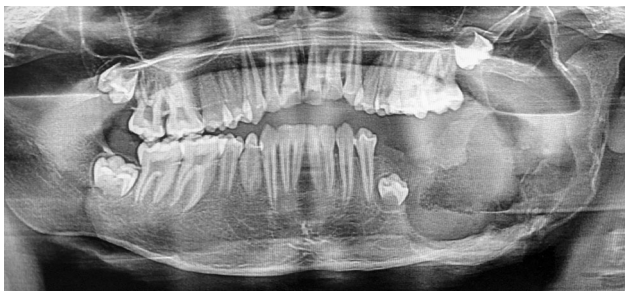


Fig. 3 Histological variety of ameloblastoma

Picture 2 .



Picture 3 .

Discussion

The treatment of intraosseous ameloblastomas is controversial, and management differs from centre to centre. In general, there are two specific surgical approaches: those who advocate a conservative approach and others who recommend that a radical approach should always be used. The conservative treatment includes an array of procedures ranging from simple curettage, enucleation alone and enucleation with peripheral ostectomy along with other adjunctive procedures including chemical cauterisation or cryosurgery. The radical approach involves marginal resection or full-thickness resection.

There has always been a vociferous demand for radical resection, significantly lowering the recurrence rate. This obviously would result in a significant deformity in the patients, most of whom are in the younger age group. Also, the inferior alveolar nerve is killed during the ostectomy resulting in permanent anaesthesia of the mental region, lower lip and teeth on the affected side. There have been attempts to preserve the inferior alveolar nerve during resection of the mandible followed by immediate bone grafting of the defect [5].

There has been a divergent view on the role of the histological variety in determining treatment based on recurrent potential. Gardner [6] has emphasised that the

histological patterns exhibited by the tumour have no bearing on the biological behaviour.

In contrast, Ueno et al. [7] have reported that the follicular type shows a multilocular appearance and showed more recurrence while the plexiform type showed unilocular appearance with lesser recurrence rates. In our study, we did not treat the pathology based on the histopathology except ameloblastic carcinoma where resection was the only option.

The treatment strategy to be adopted for ameloblastomas should be based on multiple factors like age, extent of the lesion and most importantly the availability for follow-up. Considering the locally invasive nature and extremely rare metastatic potential, treatment should be discussed based on morbidity and quality of life of the patients, noting that recurrence rate is not always the primary factor [8].

This is where the role of conservative treatment of this entity becomes an important aspect of treatment options available to the maxillofacial surgeon.

The arguments favouring adopting a more conservative approach includes the benign nature of the tumour, its slow-growing pace and the fact that it invades intertrabecular spaces of cancellous bone but does not invade compact bone but may only erode it [6]. Recurrence is limited to a small region which can be treated more easily than the original lesion.

The predominantly intraosseous nature of the pathology facilitates complete removal of residual cancellous bone using coarse surgical burs until one is sure that the compact bone is macroscopically clear. It is a fact that the architectural pattern of the tumour is such that complete removal can seldom, if ever, be achieved by enucleation alone [9]. That is why there is a need to complement the enucleation with mechanical and chemical means of removal of the surrounding bone.

The benign and slow-growing nature of the tumour may permit initial conservative management with radical salvage surgery performed on an as-needed basis if tumour recurs [10].

Pogrel and Montes [4] have pointed out that even when some mandibular cystic ameloblastomas are already very large and the cortical plates are usually very thin and even perforated, aggressive resection can cause severe deformity and dysfunction. Since it is essentially benign and rarely life threatening, it is not reasonable to reduce recurrence at the sacrifice of a patient's postoperative quality of life. The authors have thus used enucleation combined with peripheral ostectomy (Enu/PO) for the management of large mandibular cystic ameloblastomas in fourteen patients. The biomechanical integrity of the mandible was thus significantly reinforced. It is also important to preserve the periosteum, even in areas of bone perforation. Another advantage of Enu/PO is that the integrity of inferior alveolar nerve can be preserved.

Several small case studies have reported long periods of remission and potential cure after enucleation and application of Carnoy's solution. This technique has evolved from the management of keratocysts, and its use in the treatment of ameloblastomas is promising with recurrence of 10%. Haqa et al. [11] reported 27 consecutive patients with ameloblastoma of the mandible who were treated by enucleation and application of Carnoy's solution. Their argument was the strong affinity for bone and the fact that it rarely migrates into soft tissues as the periosteum forms a barrier and stops it.

Marsupialisation of an extensive lesion also seems to have a very good prognostic value as evidenced by the case report of a treatment in a 7-year-old child [12].

The conservative approach challenges a number of accepted precepts. As the preoperative histological classification is not reliable, management must be influenced by the radiological appearance. Size alone is not a defining factor, as the tumour tends to push forwards rather than progressing rapidly. Chapelle et al. [13] have also advocated that conservative treatment might be adequate initial treatment for smaller intraosseous ameloblastomas, particularly lesions that affect the mandible where the risk of spread to vital structures is small.

There have also been extensive studies on specific types of ameloblastomas. Robinson and Martinez [14] were the first to suggest that the biological behaviour of unicystic ameloblastoma makes it less aggressive than solid ameloblastoma. Similar results have been shown by Lee et al. [15]. In performing enucleation of the lesion, teeth directly related to the periphery of the tumour were extracted before proceeding with the enucleation. If the inferior alveolar nerve was exposed during the enucleation procedure, the cystic tumour was carefully stripped from the nerve which was preserved.

Till 2013, wide resection was the preferred treatment modality in our institution but the above-mentioned points then led to a change in treatment protocol thus sparing the patient of extensive loss of tissue and morbidity and therefore improving the quality of life of these young patients.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

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