Management of Ameloblastoma: Desirable Treatment Approach

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Summary

Ameloblastoma is regarded as a slow growing benign odontogenic tumor of the jaw exhibiting a locally aggressive behavior, evident by the facial disfigurement it causes. The challenges in the management of Ameloblastoma are to provide complete excision without recurrence and reconstruction of the defect with good functional and aesthetic outcome. After reviewing 10 cases (5 each, treated conservatively and surgically respectively), it has lead us to believe that radical surgical resection of Ameloblastoma followed by reconstruction of the defects is the most desirable treatment approach.

Key words: *Ameloblastoma, Enucleation, Resection, Reconstruction.*

Introduction

Odontogenic tumors are the ones that arise from odontogenic / tooth forming tissues, they present with diverse histopathological and clinical behavior. According to the definition of WHO, odontogenic tumors are defined as a locally invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma. Its behavior has been described as being benign but locally aggressive. Majority occurs in the body and ramus of the mandible, accounting for 75-80% cases and the left 20-25% are found in maxilla, predominantly in canine and premolar region, which in some cases even extend into the maxillary sinus and floor of the nose. Ameloblastoma presents with equal frequency in both sexes.

65 % occurs in 20-50 years of age with half in the 3rd and 4th decade of life. Clinically Ameloblastoma is classified into: unicystic, solid or multicystic, peripheral, and malignant. Ameloblastoma is histologically classified into a plexiform, follicular, desmoplastic, granular cell, basal cell and acanthomatous. Radiologically it presents as a unilocular or multilocular lesion, ideally referred to as "soap bubble appearance". In most cases the tumor is asymptomatic, presenting as an incidental finding on orthopantomography (Nemsadze, 1995).

Ameloblastomas are often associated with the presence of unerupted teeth (Lambrecht, et al., 2008). Symptoms include painless facial swelling and deformity, malocclusion, loose teeth, ill-fitting dentures, periodontal diseases or ulceration. Pain may be present if the swelling impinges on the vital structures. Ameloblastomas are usually treated with two therapy strategies a conservative treatment for less aggressive and smaller lesions and Radical procedures for larger lesion which later require reconstruction of the defect.

Conservative treatment is usually carried out in patients who present with smaller lesions. After confirmation of the diagnosis of Ameloblastoma based on excisional biopsy of the lesion, it is further treated by enucleation and bone curettage including the surrounding healthy bone. Precaution is taken to avoid damage to the inferior alveolar nerve while curetting the bony canal in mandibular posterior region.

Radical surgery is an operative treatment modality performed under general anesthesia, where the tumor is completely resected (segmental and marginal) with removal of blood supply, lymph nodes and also adjacent normal bone leaving a tumor free margin of 1.8 - 2cm. The large mutilating bony and soft tissue defects are reconstructed using bony grafts (iliac crest, allogenic bone material) and reconstruction plates (titanium).

Management of Ameloblastoma has always been challenging. The slow growing, locally invasive nature of the tumor along with its high recurrence rate (15-25% after radical treatment and 75-90% after conservative treatment) makes conservative treatment a questionable approach for the management of Ameloblastoma.

Owing to the recent advances in various surgical techniques like microsurgery and additive post-operative treatment modalities like radiotherapy and chemotherapy, radical surgical resection of Ameloblastoma followed by reconstruction of the defects has become the most desirable treatment approach.

Materials and Methods:

Between 2010 and 2013, 10 patients diagnosed with Ameloblastoma (8 males and 2 females) in the department of Oral and Maxillofacial Surgery were included in this study. The age group ranged from 30-60 yrs. According to the clinical and radiological data the tumors found in these patients were basically of two types, unicystic and multicystic. The tumor arose from mandibular anterior, ramus and molar region in 8 patients and the remaining 2 patients presented with tumors in maxillary canine and premolar region.

Table I. Patient demographic and clinical data

Patient	Age	Gender	Localization
1	45	Male	Maxilla, premolar region
2	42	Female	Mandible, crossing midline
3	53	Male	Mandible, anterior region
4	40	Male	Mandible, ramus area
5	38	Male	Mandible, area of the 3rd molar teeth
6	55	Male	Mandible, posterior region
7	60	Female	Mandible body
8	38	Male	Mandible, anterior region
9	36	Male	Maxillary canine region
10	52	Male	Mandible, posterior region

Attributing to the clinical, histological and radiological examinations the following treatment modalities were employed, radical surgery and Conservative treatments. Half of the cases were treated surgically and the remaining conservatively, hence dividing them equally into two study groups namely Group I (treated conservatively) and Group II (treated by radical surgery).

Figure 1. Patient with Ameloblastoma

The 5 Patients of Group I which included 2 unicystic and 3 multicystic ameloblastomas, underwent enucleation with bone curettage where the infected bone along with the tumor was removed using spoon curettage and bone ronger and the bone edges were smoothened using round diamond burr to enhance the post -operative healing.

The 5 patients of Group II which included 4 multicystic and 1 unicystic ameloblastomas, were treated by radical surgical approach. After complete resection of the involved jaw, the defects were reconstructed using iliac crest bone graft, allogenic bone material and titanium plates.

The recurrence rate in both the study groups were evaluated by clinical and radiological follow up every 6 months over a period of 3 years.

Results:

This study included 10 patients (8 males & 2 Females) in the age group of 30-60 years. The follow up period ranged from 6 months to 3 years. The particulars of the patients are presented in Table I.

The postoperative follow-up in Group I revealed unsatisfactory healing with recurrence in 4 patients (3multicystic, 1 unicystic), except for 1 patient (unicystic Ameloblastoma) where the conservative treatment was successful with uneventful healing and no recurrence. The 4 patients who presented ineffective response to conservative treatment were kept under observation and were regularly followed up.

The postoperative follow-up (clinical & radiological) in Group II showed uneventful healing without recurrence in 4 patients (3 Multicystic, 1 Unicystic) except (multicystic), where the patient presented with serious infection due to poor general health condition and smoking habit which necessitated continuation of antibiotics and drainage with regular irrigation.

Table II. Analytical results for Group I

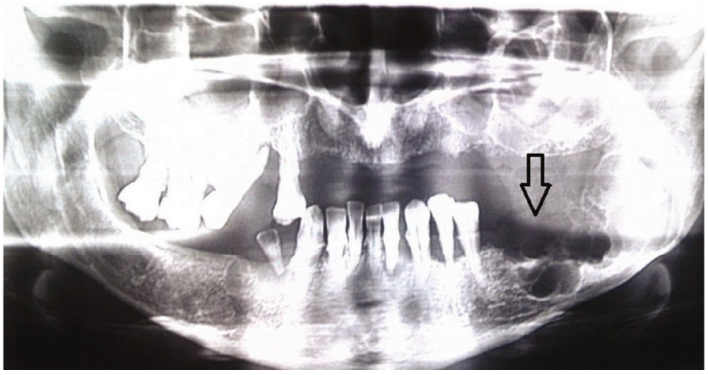
Patient	Age	Gender	Localization	Type	Results & Prognosis
1	45	Male	Maxilla	Multicystic	Ineffective
2	42	Female	Mandible	Multicystic	Ineffective
3	53	Male	Mandible	Unicystic	Very Effective
4	40	Male	Mandible	Multicystic	Ineffective
5	38	Male	Mandible	Unicystic	Ineffective

Table III. Analytical results for Group II

Patient	Age	Gender	Localization	Type	Results & Prognosis
1	55	Male	Mandible	Multicystic	Very effective
2	60	Female	Mandible	Multicystic	Effective
3	38	Male	Mandible	Unicystic	Very Effective
4	36	Male	Maxilla	Multicystic	Very Effective
5	52	Male	Mandible	Multicystic	Ineffective

The results of both the treatment modalities in the respective subgroup of patients were classified as very effective, effective and ineffective based on their postoperative healing and recurrence rates. The study Group I treated conservatively presented 4 patients with ineffective results and 1 patient with very effective result. The study Group II treated with Radical surgery and reconstruction presented 3 patients with very effective, 1 effective and 1 patient with ineffective results.

Figure 2. OPG of the patient reveals a large multilocular radiolucency (Arrow pointing to the Soap-Bubble appearance of the tumor)



Discussion

Gorlin identifies Cusack as the first person to identify Ameloblastoma in 1827. Falkson gave a detailed description in 1879. The first histopathologic description was given by Wedl (1853) who called the tumor cystosarcoma or cystosarcoma adenoids and thought that it could have arisen from the tooth bud / dental lamina. Wagstaffe (1871) gave the first histological drawing. Malassez (1885) introduced the term `adamantine epithelioma, while Derjinsky (1890) introduced the term 'adamantinoma'. However, this term has become obsolete and has to be avoided. Ivy and Churchill in 1930 encouraged the use of the term 'ameloblastoma' which is the preferred terminology till date (Punnya, 2011).

Ameloblastoma, although rare, is the most common odontogenic tumor accounting for 1% of all tumors in the head and neck region and around 11% of all odontogenic tumors (Adebiyi, Ugboko, Omoniyi-Esan, Ndukwe, & Oginni, 2006). In the newest classification by the World Health Organization, variants of ameloblastoma are categorized on the basis of characteristics, such as the age at presentation, location in the body, imagining features, clinical behavior and prognosis. Thus, the plural term ameloblastomas is used to describe this family of diseases (Reichart, Philipsen, & Sciubba, 2006). Ameloblastomas are classified as either extraosseous (peripheral) or intraosseous. Peripheral ameloblastomas manifest as a sessile or pedunculated slow-growing mass that is confined to the gingiva or alveolar mucosa with no involvement of underlying bone. Intraosseous ameloblastomas arise in the jaw and are further classified as unicystic, desmoplastic and mixed cystic and solid. "The mixed cystic and solid form demonstrates more aggressive behavior and is more likely to recur than unicystic and desmoplastic ameloblastomas." Larsson and Almeren report the incidence of ameloblastoma in Sweden as 0.3 cases per million per year. The average age of patients with intraosseous ameloblastoma has been reported to be 39 years and appears equal frequency between sexes, although a higher frequency in females than in males has been described." Our study groups had findings similar to that of Fregnari et a1 (Fregnani, et al., 2010), 80% of the tumors were located in mandible, 70% were located in the area of molars or the ascending ramus, 20% in the premolar region and 10% in the anterior region.5 About 10-15% of ameloblastomas are associated with a nonerupted tooth (Lambrecht, et al., 2008).

Ameloblastoma was known for its high recurrence rate if excision was incomplete. Therefore the treatment of choice is surgical excision with wide free margins. The traditional approach for a mandibulectomy is through a lip -splitting incision and though it has the disadvantage of post-operative morbidity; it gives a better exposure for complete tumor removal. Some authors such as Derderian et al (Derderian, Gurtner, & McCarthy, 2004) use a less invasive incision which avoid troublesome outcome of the lip-splitting. They utilize a Risdon incision and this was combined with intra-oral incision which gives a less post-operative morbidity and more cosmetic outcome (Derderian, Gurtner, & McCarthy, 2004). A new technique of removal of large ameloblastoma with immediate reconstruction by using only an intra-oral incision. It has the advantages of removing and repositioning of the mandible intra-orally and therefore allows removal of the lesion and reconstruction procedure to be done simultaneously. Facial scar and damage to the marginal mandibular nerve that innervates the lips can also be avoided via this technique.

However, extensive tumors require a more radical approach. The amount of resection is variable and depends on the site and extension of the tumor. All the Patients included in our study presented with locally advanced tumors, already infiltrating the surrounding soft tissue. According to our study, the results of conservative treatment in these cases were not satisfactory as they resulted in local recurrent tumors making further surgical treatment even more complicated resulting in cosmetic and speech deficits.

On the other hand radical surgical treatment modalities were successful in complete resection of the tumors in most of the patients. Although the challenging aspect of this treatment modality is the reconstruction of post-surgical defects. These defects have to be managed decorously for the success of the treatment restoring adequate functional and aesthetic outcome assuring good mental

and social wellbeing of the patient. Postsurgical defects in the maxillary region predispose the patient to hypernasal speech, impaired masticatory function, fluid leakage into the nasal cavity, and various degrees of cosmetic deformity. Mandibular resection can prove to be devastating to mastication, deglutition, phonation, and oral competence. Moreover, the mandible frames the lower third of the face and represents a major component of the human appearance. Satisfactory reconstruction of complex jaw defects, especially in a single-step procedure, is therefore a surgical challenge. For benign tumors, the bone grafts have become a reliable source during the last few years in osseous reconstruction. The fibula, scapula and iliac crest are the commonly chosen donor sites to reconstruct mandibular or maxillary defects. For reconstruction of defects in the mandible we preferred iliac crest bone grafts, as it provides a good quality of bone in sufficient amount.

Figure 3. Intraoperative view of the Mandible with Ameloblastoma

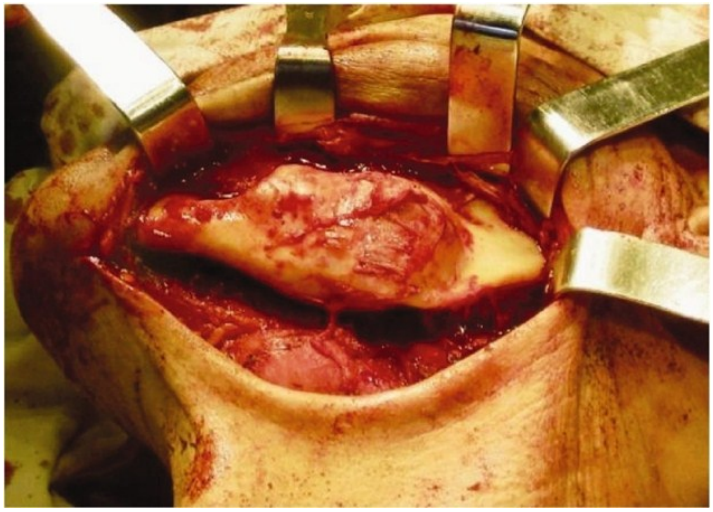
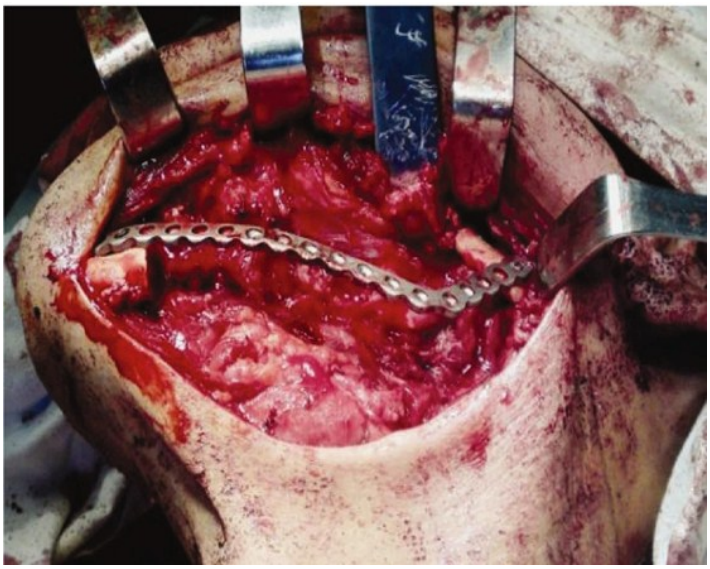


Figure 4. Resected Part of the Mandible

Figure 5. The defect fixed with titanium plate and screws



In our follow-up regime, patients were scheduled for clinical and radiological examination half yearly for the first 3 years followed by once a year there after. We suggest a long follow-up period for at least 10 years as recurrence may also appear years after primary surgery.

In conclusion, Radical surgical resection of ameloblastoma followed by reconstruction of the defects is the most desirable treatment approach. The challenge in the management of large ameloblastoma of the mandible is not only to excise the tumor completely in order to prevent recurrence but also to provide the best aesthetic and functional outcome through various reconstruction methods.

The above results establish the effectiveness of conservative treatment to be 20%, in contrary to the radical surgical treatment with 80% effectiveness.

Figure 6. Postoperative X-ray

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