Case Report I

S R Gunawardane and A M Attygalla

ABSTRACT
Considering all swellings of the oral cavity, 9% are odontogenic tumors and among them, ameloblastoma accounts for 1% of lesions. Surgical resection with wide margins (1 cm from the radiological margin) has been considered as the principle treatment of solid multicystic ameloblastoma. However, in most cases, the lesion may be very extensive at the time of presentation because the tumor is painless and shows slow and expansive growth. The resection of the jaw including condyle and the anterior region in a growing young patient is associated with a number of complications such as loss of bony support of the jaw, deformity, dysfunction and psychological disturbances even after reconstruction. An alternative conservative surgical procedure called the “Dredging Method” can eliminate the tumor while restoring the normal contour and function of the jaw.
Key Words: Ameloblastoma, Dredging method, Conservative management

INTRODUCTION
Odontogenic tumors are a diverse group of lesions with a wide range of histopathological types and clinical behavior. Considering all swellings of the oral cavity, 9% are odontogenic tumors and among them, ameloblastoma accounts for 1% of lesions.[1] According to the World Health Organization, it is defined as a locally invasive polymorphic neoplasm that often exhibit a follicular or plexiform pattern within a fibrous stroma. Its behavior has been described as being locally aggressive. In 20% of the cases the tumor can be found in the maxilla, predominantly in the canine or molar region. When considering the mandible, 70% are located in the molar region or the ascending ramus, 20% are found in the premolar region and 10% are in the anterior part. Ameloblastomas occur with no gender predilection.[2] The predominant age range is between the first and the seventh decade of life with a peak frequency in the fourth decade. Clinically, they can be classified into 4 groups: unicystic, solid or multicystic, peripheral, and malignant. The unicystic type usually appears as a “cystic” lesion with either an intraluminal or a mural proliferation of the cystic lining. Radiographically, it may resemble a well-circumscribed radiolucency. Multicystic type can infiltrate into the adjacent tissue with the potential ability to recur and even metastasize. Radiographically, the appearance is generally unilocular or multilocular. Most common site for the peripheral type is the alveolar mucosa. It is a soft-tissue variant of an ameloblastoma, sometimes may involve the underlying bone. The malignant ameloblastoma is a rare entity with metastatic potential but still maintains its classical microscopic features.[3,4] According to histopathological classification, ameloblastomas subdivides into follicular, plexiform, basal cell, acanthomatous, and granular types. Mostly the tumor is asymptomatic. The most common symptoms include facial swelling and disfigurement, pain, malocclusion, loosening and mobility of teeth, ill-fitting dentures, periodontal diseases, ulceration of oral mucosa, oroantral fistulas and upper airway obstruction.

Dr. S R Gunawardane (BDS) (Correspondence) Temporary Lecturer, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, University of Peradeniya, sumithgunawardane7@gmail.com
Dr. A M Attygalla (BDS, MD) Senior Lecturer and Consultant Oral & Maxillofacial Surgeon, Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, University of Peradeniya, manjuatty@yahoo.com
Two therapeutic strategies have been discussed in the literature: conservative modality and radical procedures. While smaller less aggressive lesions are treated by a less aggressive approach; larger, aggressive lesions require radical surgical intervention resulting in large defects making reconstruction difficult.⁶

Management of ameloblastoma is controversial since the biological behavior of this disease is unique in growing slow and constitutes a locally invasive tumor with a high chance of recurrence. The recurrence rates are reportedly as high as 15-25% after radical treatment while 75-90% persists after conservative treatment. Therefore, wide resection of the jaw in accordance with the management of malignant tumors is usually recommended.⁷

THE DREDGING METHOD
Surgical resection with wide margins (1 cm from the radiological margin) has been considered as the principal treatment of solid multicystic ameloblastoma. However, in most cases, the lesion may be very extensive at the time of presentation because the tumor is painless and shows a slow and expansive growth. The resection of jaw including condyle and the anterior region in a growing young patient is associated with a number of complications such as loss of bony support of the jaw, deformity, dysfunction and psychological disturbances even after reconstruction. An alternative conservative surgical procedure the “Dredging Method” is a procedure which can eliminate the tumor while restoring the normal contour and function of the jaw.⁸

The term dredging comes from soil science where dredging is defined as an excavation activity usually carried out underwater, in seas or freshwater areas with the aim of collecting bottom sediments and scavenging of them at a different location. [⁹]

The “Dredging Method in the management of ameloblastoma” is a maiden conservative surgical procedure wherein after deflation and enucleation or only enucleation, repeated dredging is applied to accelerate osteogenesis by eliminating the scar tissue from the bony cavity. Deflation is used in large cystic lesions, where part of the cystic wall, overlying bony covering and mucoperiosteum are removed in order to reduce the intracystic pressure and facilitate the formation of a demarcated bony outline. Enucleation is done following the formation of demarcated bony outline; on the other hand enucleation can be done directly in solid ameloblastoma. After enucleation, the tumor is eliminated completely along with a part of the surrounding healthy bone, and next the bony cavity is kept open. Then the procedure is followed by repeated regimes of dredging the scar tissue that is formed in the bony cavity. Dredging is applied within 2-3 months interval to accelerate new bone formation and eliminate tumor cell nests. Histopathological examination of all specimens are essential to ensure elimination of residual tumor cells and prevention of recurrence. The follow up in the “Dredging Method” begins when the tumor cells cannot be identified in microscopic examination of the tissues removed by 2 consecutive dredging procedures. Long term and regular follow up is an essential aspect of the treatment regime. [10]

CASE REPORT
A 23 years old female patient presented to the Department of Oral & Maxillofacial Surgery, Faculty of Dental Sciences, University of Peradeniya in the 2006 with a gradually increasing swelling of the right side mandible of two weeks duration. The patient was referred from a regional dental hospital. The patient had a history of asthma (not on medication) and was taking Amoxicillin as prescribed by the referring clinician. On examination soft swelling was noted in the 46-47 region extending to the buccal sulcus. Lateral oblique radiograph and Dental panoramic tomographic radiograph confirmed the swelling as a cystic lesion in relation to 46-48 region. An exploration biopsy was performed
under general anesthesia and diagnosis made as anon-specified cystic lesion on the right side mandible. Decompression of the lesion was initiated. Due to the several episodic recurrence of the cystic lesion decompression had to be repeated on several occasions in 2007, 2008 and 2009. Repeated biopsies were done and all of them were repeated as anonspecific cystic lesion of the mandible. The decompression was continued until the lesion completely disappeared both clinically and radiologically.

Again the patient presented in the year 2010 with a recurrent lesion of the same site and second exploration biopsy was performed. The biopsy revealed that the lesion as a follicular ameloblastoma. An ultrasound scan of the neck was performed and it showed multiple reactive lymph nodes in left side submandibular region. Enucleation of the lesion was done in June 2010 and periodic irrigation were continued until the lesion was completely healed.

The tumour recurred in the year 2013 and third exploration biopsy confirmed it as solid multicystic ameloblastoma (follicular variant). As the recurrence nature and multicystic behavior of the lesion, the most common treatment option was the resection of the right side mandible and thereby removing the entire lesion. Rather than going for a resection of the mandible, it was planned to initiate dredging as the most conservative management for this kind of lesion (Figure 1). The dredging was initiated in December 2014 and repeated in March 2015, July 2015, October 2015 and February 2016 respectively. The biopsies were positive for tumour cells on March 2015, July 2015 and October 2015 with clear radiological evidence for reduction of tumor size. The biopsy report of February 2016 was the first which came as a tumour free and the consecutive biopsy obtained from the May 2016 (Figure 2) also confirmed the tumour free status. The dredging method was successfully concluded and the patient was advised to attend regular follow-ups.

**DISCUSSION**

When planning treatment of ameloblastoma, it is important to understand the growth characteristics and to remove the full extent of the tumor, including the surrounding tissues. Otherwise, the remaining tumor cells may lead to multiple morbidities of recurrence. Recent advancements in the understanding of the biological behaviors of ameloblastoma have revealed that unicystic lesions are well localized by the fibrous capsule of the cyst, with a few tumors broaching peripheral tissues, whereas multicystic and solid lesions are characterized by an aggressive infiltration to adjacent tissue. Gardner discussed the treatment of ameloblastoma on the basis of pathological and anatomical considerations. He stated that the recommended treatment for solid and multicystic ameloblastoma was radical treatment, whereas unicystic ameloblastoma was usually cured by curettage. [11]

Two therapeutic strategies are mentioned in literature: a conservative way of treatment and
radical procedures. Non-radical surgical procedures like enucleation and curettage, combined with liquid nitrogen spray cryosurgery, or just drilling of the peri-lesional bone are mentioned to be useful in unicystic ameloblastomas, especially in children and young patients. Payne et al 2015 and Larrañaga et al 2015 showed high rates of recurrence of ameloblastoma after conservative treatment protocols and therefore recommend radical surgical treatment. Larrañaga et al 2015 suggests a “rational radical conservative” resection of the mandible with preservation of the lower border of the mandible to maintain the continuity of the lower jaw and the facial contours when there is no cortical perforation.

Sehdev et al 1974, reported recurrence after the conservative approach (curettage) in more than 90% of 92 ameloblastomas. Shatkin and Hoffmeister 1965 reported that 86% of 20 mandibular ameloblastomas recurred after curettage compared with a 14% recurrence rate after en bloc resection. Ackermann et al 1988 have reported a series of 57 ameloblastomas in which they found a 52% rate of recurrence in patients treated conservatively and a 25% rate of recurrence in patients with primary tumor treated by the radical approach. However, extensive tumors require a more radical approach. The amount of resection is variable and depends on the site and extension of the tumor. This patient also had undergone decompression, enucleation like treatment methods for a long duration. However there were three recurrences so far. Considering the high recurrence rate, wide resection of the jaw is usually the recommended treatment for ameloblastoma. However, radical surgery often means that the patients have serious complications including facial deformity, masticatory dysfunction, and abnormal jaw movement. Considering the characteristics of ameloblastoma as a locally invasive but slow growing and rare metastasizing benign tumor, the priority of the treatment method should be discussed from the points of morbidity and quality of life of the patients, noting that the recurrence rate is not always the primary factor. Postsurgical defects in the maxillary region predispose the patient to hypernasal speech, fluid leakage into the nasal cavity, impaired masticatory function, and in some patients, various degrees of cosmetic deformity. Mandibular resection can also prove 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 donorsites to reconstruct mandibular or maxillary defects.

The contour of the face and oral cavity is directly related to the function and facial aesthetics. So, treatment of diseases of the oral cavity becomes inadequate if it causes deformity of face. Deformity of the oral cavity causes functional inconvenience, aesthetic dissatisfaction and mental agony. So, the purpose should be correction of disorder as well as to restore normal contour and function of the jaw. Considerations should be given to the age of the patient, site, nature, extension of lesion. The Dredging Method is considered to fulfill these purposes. It is seen after deflation and enucleation that the tumor cells are identified in the scar tissue within the bony cavity which is the cause of recurrence. So the scar tissue should be dredged out repeatedly to prevent the recurrence as well as to accelerate new bone formation. It is reported in the literature that, there is a very low recurrence by the dredging technique as compared to other conservative techniques. Considering the advantages of the dredging method, this patient has shown positive improvements so far.

CONCLUSIONS
Ameloblastoma has a high rate of local recurrence if it is not adequately removed. In our opinion, even though radical surgical resection of

Ameloblastoma is the treatment of choice, and dredging could be used in order to reduce the post-surgical complication and to achieve better quality of life in the long run. However, the chances of recurrence in this method had to be surveyed more in order to gain a better outcome.

REFERENCES


