Management of Plexiform Ameloblastoma: A Case Report

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ABSTRACT
Ameloblastoma is a slow growing benign tumour of the jaw and patients usually present late after the tumour achieves considerable size to cause facial disfigurement. Diagnosis made mainly from tissue biopsy and characteristic findings on plain X-rays and CT scan, they assist in differentiating between types of ameloblastoma. The challenges in the management of this tumour are to provide complete excision as recurrence may occur in incomplete removal and also to reconstruct the bony defect in order to give reasonable cosmetic and functional outcome to the patient.

Key words: Ameloblastoma, Reconstruction

INTRODUCTION
The ameloblastoma is a slow growing, benign but locally invasive neoplasm. It may arise from the enamel organ, the dental follicle, the periodontal ligaments and the lining of odontogenic cysts (1). Ameloblastomas are the most common odontogenic tumors. They are usually diagnosed in the fourth and fifth decades. About 80% of the tumors occur in the mandible (2), of which three quarters are in the molar region and the ascending ramus. A 20% occur in premolar region and rest in incisor region. A typical intraosseous ameloblastoma is locally invasive with islands of tumor infiltrating cancellous marrow spaces without causing bone resorption. As a consequence simple curettage is not sufficient and recurrence rates of 55–90% have been reported with enucleation (3). The recommended treatment is radical resection of the affected part of the jaw.

Various methods of reconstruction of mandible have been described. Replacement with cancellous bone and a tray, fibula graft, rib graft, spine of scapula, and iliac crest graft are few of them (3-5).

CASE REPORT
A 30-year-old female patient reported to our maxillofacial unit with complaint of swelling on right side of face since last 10 months. A history of progressive increase in size, not associated with pain was elicited. There was no history of ulceration, discharge, bleeding, or difficulty in opening the mouth.

On extraoral examination there was a uniform ovoid swelling of approxi

![Figure 1a: Preoperative view of the patient. The swelling on the right mandible was visible](image-url)
approximately 4 x 5 cm over lower jaw, extending from the zygomatic region to the inferior border of mandible superoinferiorly, and from the corner of mouth to the angle of mandible anteroposteriorly. It was nontender, bony hard in consistency, nonpulsatile and neither compressible. There was no sensory or motor deficit on right side of face. There was no cervical lymphadenopathy (Figure 1a).

Examination of oral cavity revealed poor oral hygiene with right lower second molar missing and ulceration present over right buccal mucosa. There was mild right lateral bulge in floor of mouth that was again bony hard. Routine biochemical and hematological investigations were within normal limits.

The panoramic view of the jaw revealed multicystic lesion in the body and ramus of the right mandible extending up to the sigmoid notch and condylar neck. Inferior displacement of right mandibular 2nd molar and root resorption of the right 1st molar. Inferior alveolar canal displaced caudally (Figure 1b).

CT scan showed a large multilobulated bony cystic lesion, extending from angle of mandible inferiorly and involving the coronoid and condyle completely superiorly. Anteroposteriorly completely involving the ramus of the mandible and invade in to soft tissue (Figure 1c).

Biopsy of the mass suggested the diagnosis of benign odontogenic tumor with ameloblastic differentiation.

Under general anesthesia and nasoendotracheal intubation and all aseptic precautions, tumor mass was exposed buccally and lingually via lip splitting incision. After extraction of right lower lateral incisor, osteotomy cut was placed and completed buccally and lingually. Whole of right hemimandible exposed till condyle and coronoid process above. Right tempomandibular joint disarticulated. The expansile swelling was removed and sent for histopathology.

Defect was reconstructed by using AO titanium reconstruction plate with condylar prosthesis and non-vascularized iliac bone graft was secured to the reconstruction plate by using titanium screws (Figure 2). Drain was secured and closure was done in layers. Antibiotics, analgesics and anti-inflammatory drugs were given postoperatively. Extraoral sutures were removed on 7th postoperative day. Patient has been kept under periodic follow up since then. Postoperative patient had no complaints in chewing, swallowing or speech articulation. Mouth opening was in normal limit and with no recurrence in one year follow up (Figure 3a and 3b).

Post operative histopathological examination revealed plexiform ameloblastoma.

Figure 3a: Postoperative view of the patient after one year.

DISCUSSION

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures (6). It is the most common odontogenic tumor although it represents only about 1% of tumors and cysts of the jaws (7).

In the mandible (80% of ameloblastomas), 70% are located in the area of the molars or the ascending ramus, 20% in the premolar region, and 10% in the anterior region (8). About 10-15% of ameloblastomas are associated with a non-erupted tooth. In the present case, a large plexiform ameloblastoma found in the ascending ramus and molar region of the mandible and it was not
associated with a non-erupted tooth.

Ameloblastoma appears equal frequency between sexes, although a higher frequency in females than in males has been described (6). In our case, the patient was female and was in third decade of her life.

Clinically, it frequently manifests as a painless swelling, which can be accompanied by facial deformity, malocclusion, ulceration and periodontal diseases. In our case, clinical examination revealed a large, expansive mass in the ascending ramus and molar region of the mandible. The swelling was hard, painless to palpation and covered by normal mucosa.

Histologically, ameloblastoma is characterized by the proliferation of epithelial cells arranged in a collagenous fibrous connective tissue stroma of conjunctive vascular tissue in locally invading structures that resemble the enamel organ at different stages of differentiation (6). The tumor found in our patient was an ameloblastoma of the plexiform type. The term “plexiform” refers to the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern.

When the results of treatment of ameloblastomas are assessed, several factors are important. First, long term follow up is essential because this neoplasm has the capacity for continued growth. Successful treatment in this context may be defined as treatment that renders an acceptable prognosis, causes minimal disfigurement, and is appropriate on the age and general health of the patient and on the size, location and duration of the tumor. In our case surgical resection of large sized ameloblastoma was done to prevent recurrence and mandible stabilised and reconstructed with AO titanium reconstruction plate with condylar prosthesis and non-vascularized iliac crest graft was harvested.

In conclusion, en-bloc tumour resection reduces the chance of tumour recurrence but resulted in large mutilating bony and soft tissue defects as indicated by our experience and also in many other series. The challenge in the management of large ameloblastoma of the mandible is not only to excise the tumour completely in order to prevent recurrence but also to provide the best reconstruction method.

REFERENCES