

**A LARGE AMELOBLASTOMA OF THE MANDIBLE: A CASE REPORT**Anuj Bhargava<sup>1</sup>, Smita Soni<sup>2</sup>, Amit Tyagi<sup>3</sup>**ABSTRACT**

Ameloblastoma is a slow growing, and locally aggressive tumor with high recurrence rates. The tumor can assume large sizes. It arises from epithelium of the dental lamina. Clinically the tumor can present itself as swelling which are generally asymptomatic. Histologically there are many variants. Radiographically the tumor can occur either as multilocular radioluscent lesion giving a peculiar honey comb appearance or as a unicystic variety. There are many treatment options available which range from conservative treatment of curettage, enucleation to radical surgical approaches of wide margin excision. Radical treatment approaches have the advantage of lowering the recurrence rates but at the same time pose extremely difficult challenges of reconstruction of the surgical defects. We are reporting a case of a 20 year old young female diagnosed with a large multicystic ameloblastoma of the mandible in which wide margin surgical excision of the tumor by segmental resection of the left hemimandible was performed with spanning of the bony defect with titanium reconstruction plates to achieve a favorable aesthetic and functional outcome for the patient.

**Keywords:** Ameloblastoma, Reconstruction, 3D CBCT

**Key Message:** Ameloblastoma is an anatomically benign, clinically persistent and locally aggressive tumor with high recurrence rates. Radiographically it may present as a multilocular radioluscent lesion with characteristic "soap bubble or honey comb appearance" or as a unilocular lesion. Conservative approaches of treatment include curettage and enucleation with reported high recurrence rates. Radical treatment involves wide margin excisions of the tumor mass. Reconstruction of the resected defect is a challenge for the surgeon to provide a favorable functional and aesthetic outcome to the patient.

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Ameloblastoma is a rare benign true neoplasm of odontogenic origin. It is derived from the word of English literature "Amel" which means enamel. In Greek "Blastos" refers to germ[1]. Ameloblastoma is known for its locally invasive and aggressive character. It has a strong tendency to recur which is

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well evidenced by its high recurrence rates[2].

This odontogenic tumor has been considered to be of varied origin by many authorities. Thus the tumor might be conceivably thought to be derived from cell rest of the enamel organ, either remnants of dental lamina or remnants of Hertwig's sheaths, epithelial rest of Malassez, epithelium of odontogenic cysts, particularly the dentigerous or the odontomas, disturbance of the developing enamel organ, basal cells of the surface epithelium of the jaws or the heterotopic epithelium in other parts of the body, especially the pituitary gland[3].

In 1827 Cusack was the first person to describe this tumor[4]. The name "Admantinoma" was introduced by Malassez in 1885, which nowadays is used to describe a rare type of bone cancer[5]. Churchill coined the term "Ameloblastoma" in 1930[6]. According to WHO the tumor is a benign, but locally invasive neoplasm that often has diverse histologic patterns[7].

This group of tumors comprise of various histopathological types and clinical behavior[8]. Ameloblastoma accounts for about 1% of all the cyst and tumors of the jaws and 18% of the different odontogenic tumors[9].

The mandible has five times higher occurrence of the tumor as compared to the maxilla[10]. The average age of occurrence as reported in literature is 38.9 years[11]. It occurs with equal frequency in both sexes[12]. If we review the literature, in 52% of the cases it occurred in men, 48% in women[11]. Small and Waldron[11] have reported that it is the molar region where 47% of Maxillary Ameloblastomas occur, a lesser percentage of 33% occur in the antrum and floor of the nose, only 9% occur in both the premolar and canine regions. 2% occurrence was noted in the palatal region of the maxilla according to their study. In the mandible most common site is molar and ascending ramus region accounting for 39%, and 16% occurred in molar premolar region and 9% in the anterior region[13].

Clinically the tumor can be classified in four distinct types: unicystic, solid or multicystic, peripheral and malignant. The unicystic type usually presents as a "cystic" lesion with either an intraluminal or an intramural proliferation of the cystic lining[14]. Radiographically, it is seen as a well-circumscribed slow-growing radioluscent lesion. Multicystic Ameloblastomas can show infiltration into the neighbouring tissues with ability to recur. They can sometimes show metastasis. Radiographically, it

may appear as a unilocular or multilocular lesion[15]. Peripheral ameloblastoma is a soft tissue variant of ameloblastoma which mostly presents in the alveolar mucoas. Although this lesion can also involve the underlying bone[16].

Malignant Ameloblastoma although a rare type is defined as an ameloblastoma which has already undergone malignant metastases but still has its classical histological microscopic features[17]. Other histological variants have also been described for ameloblastoma like follicular, plexiform, basal, granular and acanthomatous[18]. Desmoplastic Ameloblastoma is a rare variant which has been described in The World Health Organization(WHO) classification of odontogenic tumors lately[19].

Clinically, ameloblastomas appears as an aggressive odontogenic tumor, often asymptomatic and slow growing, with no sign of swelling. It can sometimes cause symptoms such as swelling, dental malocclusion, pain, paresthesia of the affected area[20]. It spreads by forming pseudopods in marrow spaces without concomitant resorption of the trabecular bone. Because of this, the tumor margins are not clearly seen on radiographs or during surgery and the tumor frequently recurs after inadequate surgical removal[21,22]. The appearance of septae on the radiograph usually represents differential resorption of the cortical plate by the tumor and not actual separation of tumor portions[23]. Because of its slow growth, recurrence of ameloblastoma generally present many years and decades after primary surgery[22]. When treated inadequately, malignant development is a possibility[20].

In most cases ameloblastoma has a characteristic but not diagnostic radiographic appearance[21]. The neoplasm usually appears as a unilocular radioluscent area or a multilocular radioluscent area with honey comb appearance[20,21]. Adjacent tooth roots might show evidence of resorption[21]. The tumor in many cases might be accompanied with an unerupted tooth, most commonly a mandibular third molar[24].

Treatment of mandibular ameloblastomas continues to be controversial. It can change with clinicoradiographic variant, anatomic location and clinical behavior of the tumor[25]. Treatment consists of wide resection, enucleation and curettage [22,26]. Rates of recurrence may be as high as 15 % to 25% after radical treatment and 75% to 90% after conservative treatment[26]. According to Reichart PA, Philipsen HP[13] and associates the rate of recurrence demonstrated were 17.7% for en bloc resection to 34.7% for conservative therapy.

The aim of this article is to present a case of a large mandibular ameloblastoma which was treated with radical treatment approach of segmental resection of the left hemimandible followed by reconstruction with Titanium plates to achieve a partially favorable aesthetic and functional outcome. The article briefly elaborates the clinical and histological types of the tumor

along with the recurrence rates associated with the tumor because of its unique biologic behavior. The article also discusses the importance of reconstruction especially in cases of large tumor resections.

### CASE REPORT

A 20 year old young female patient [Figure.1] reported to the outpatient department of our institute around one year back with a chief complaint of slowly increasing large painless swelling over left side of the lower jaw since last one year of her first visit to the institute. There were no signs of difficulty in deglutition or breathing. No underlying significant medical history was elicited.

Physical examination revealed a large non tender, bony hard 6cmX 6cm asymptomatic well demarcated swelling anteriorly extending from left angle of the mandible to the left corner of the mouth. The swelling extended superiorly from the left malar region inferiorly into the left hyoid bone region of the neck causing gross facial disfigurement. On palpation regional cervical lymph nodes were non tender and not enlarged. There was no evidence of any neurosensory deficit especially along the distribution of the trigeminal nerve.

The swelling was non pulsatile with normal overlying skin color and texture. Few dilated superficial veins were prominent over the swelling. Intraoral examination revealed fair oral hygiene with no signs of trismus. There was gross obliteration of the left mandibular buccal vestibule. Grade II mobility was elicited with the lower left 3rd molar. The overlying mucosa over the swelling was intact on manual palpation. The swelling intra-orally extended from the left lower canine region to the left retromolar region anteroposteriorly [Figure.2] Tongue movements and sensation were noted and appeared to be normal.

Lingual cortical expansion was palpable on the left side of the mandible. Buccal cortex was also enormously expanded. Routine blood examination, chest x-ray and other vital organ investiga-

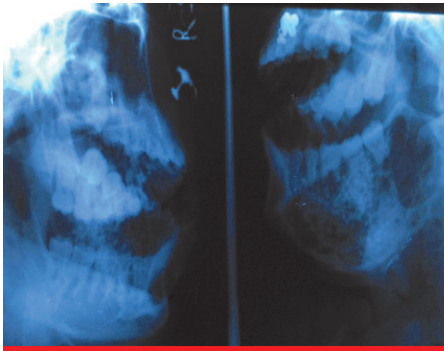


Fig.1:Pre-Operative View



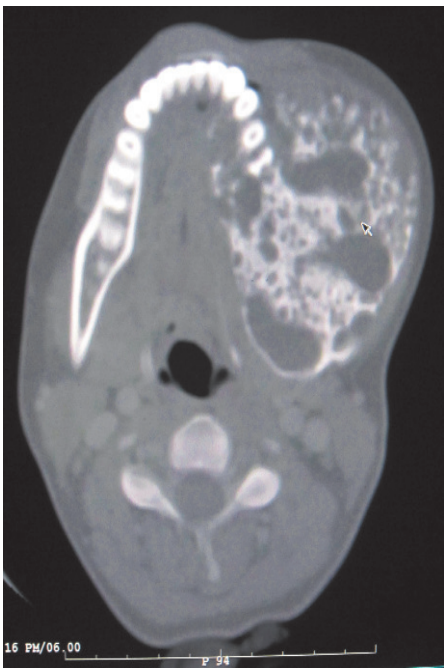
Fig.2:Intra Oral View





**Fig.3:Preoperative Left & Right Lat.Oblique 30 Degree View**

not available at our centre. A plain and contrast CT scan of the mandible with 4mm collimation was advised which revealed



**Fig.4:CT Scan Mandible**

[Figure.4] multiloculated expansile lytic lesion (7.3 cm X 6.2 cm X 7.3cm) showing multiple internal osseous trabeculae giving honey comb appearance involving the left body of the mandible and the ramus. There was cortical thinning, with multiple areas of cortical breach and adjacent small soft tissue component. Left side of the condyle and Temporomandibular Joint were free of the mass. Differential diagnosis as per the radiographic report included odontogenic fibroma, odontogenic myxoma, fibroma, ameloblastoma and central giant cell granuloma. An intra-oral aspiration biopsy was also performed which revealed yellowish straw colored fluid.

Although biopsy results did not show any specific features of ameloblastoma, the clinical presentation, history along with radiographic diagnosis was quite suggestive of ameloblastoma. The patient was planned for wide margin surgical excision of the tumor mass under general anesthesia. Nasotracheal intubation was performed and Erisch arch bars were placed on the contralateral side. The tumor site was reached by the traditional lip splitting and the submandibular incisions. Soft tissue dissection was performed carefully and the skin flaps were raised, marginal mandibular nerve was protected to expose the tumor mass.

Intraoperatively the tumor was found to be around 7cm X 6cm in size [Figure.5] with anteroposterior extensions from the left

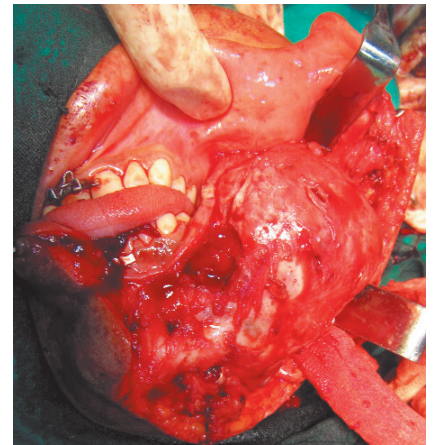
tions were also performed which were also normal.

Left and Right Lateral oblique 30 Degree views [Figure.3] along with PosteroAnterior and AnteroPosterior view of the mandible were also advised. Orthopantograph facility is not available at our centre. A plain and contrast CT scan of the mandible with 4mm collimation was advised which revealed [Figure.4] multiloculated expansile lytic lesion (7.3 cm X 6.2 cm X 7.3cm) showing multiple internal osseous trabeculae giving honey comb appearance involving the left body of the mandible and the ramus. There was cortical thinning, with multiple areas of cortical breach and adjacent small soft tissue component. Left side of the condyle and Temporomandibular Joint were free of the mass. Differential diagnosis as per the

incisal region up to the left ascending rami of the mandible sparing the coronoid and the condyle. Segmental resection of the left hemimandible was done from the left incisal region to the left ascending ramus of the mandible leaving the coronoid process and the bony chunk of subcondylar region of the mandible. The tumor was freed from all its soft tissue attachments and removed in totality [Figure.6].

The specimen was sent for histopathologic diagnosis [Figure.7]. The bony defect created by the resected mass was spanned with titanium reconstruction plates [Figure.8] taking due consideration of the occlusion of the contralateral side which was previously secured with arch bars. Hemostasis was achieved. Surgical drains were placed. Wound closure was done in layers. Post operative recovery was uneventful. The patient was kept on intermaxillary fixation from the second post operative day. The extraoral sutures were removed after 1 week. The patient was also kept on ryles tube feeding for 2 weeks and later instructed to start liquid diet orally. Patient was discharged after 2 weeks.

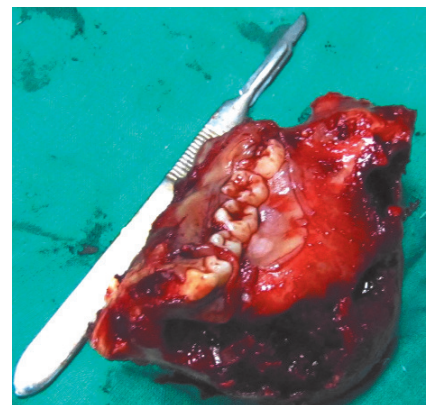
Post operative histopathologic report suggested of bone tissue with tumor composed of growth and islands of odontogenic epithelium with peripheral pal-



**Fig.5:Intraoperative Tumor measuring 7cm X 6 cm in Size**



**Fig.6:Tumor mass Resected**



**Fig.7:Tumor Specimen**



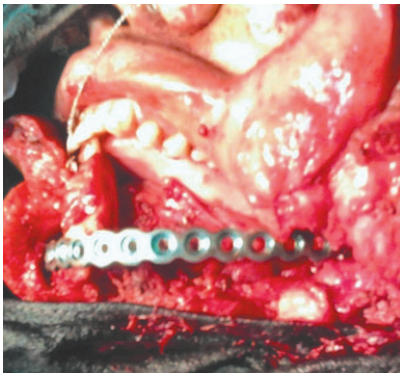


Fig.8:Boney Defect Spanned by Ti Reconstruction Plates

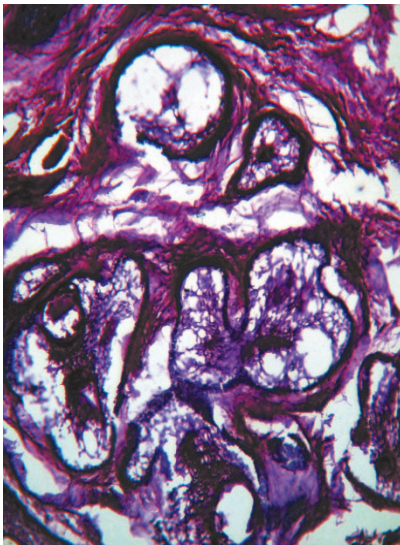


Fig.9:Histologic Specimen Suggestive of Ameloblastoma of Follicular Type



Fig.10:Post Operative View

lisading and central reticular pattern. However there was no cytological atypia or increased mitotic activity. Rest of the tissue was reported free from any tumor deposits. Histo-logically the specimen was suggestive of ameloblastoma of follicular type [Figure.9].

Patient was kept on regular follow up visits and Intermaxillary fixation was removed after the fourth week [Figure.10]. A post operative 3D Cone Beam CT scan was also advised to the patient [Figure.11]. Patient did not complain of any malocclusion or difficulty in deglutition on subsequent follow-ups. Patients voice was also normal. She has been regularly visiting the department and till now there have been no signs of any discomfort or recurrence.

## DISCUSSION

Ameloblastoma is the second most common odontogenic neoplasm occurring in the oral cavity after odontoma[2]. In our report the patient is a 20 years old female who presented with a large ameloblastoma of the left mandible. According to the study conducted by Reichert PA and Philipsen HP[13] in mandible the most common site of occurrence is molar and ascending ramus region accounting for 39% .In

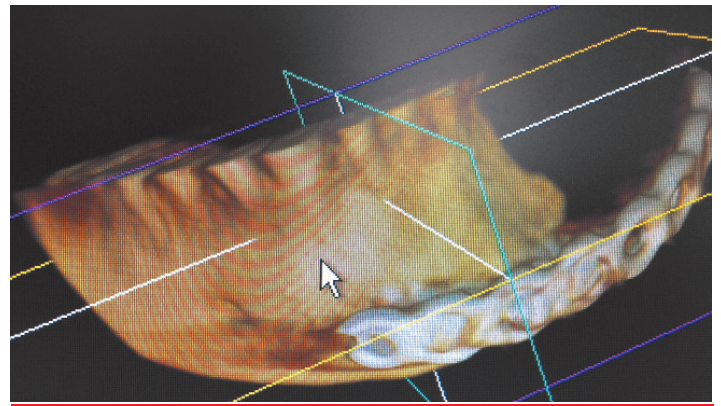


Fig.11:Post operative CBCT

our case the site of occurrence of the tumor was also the molar and ascending ramus region.

Ameloblastomas can range from small to very large sizes causing gross facial asymmetry and disfigurement. Apart from causing aesthetic problems they also cause functional disturbances like malocclusion, loosening of teeth, paresthesias and pathologic fractures. Tumor can cause expansion of cortical plates along with local bone invasion. The tumour can present as a slowly progressive painless mass which may lead to aforesaid deficiencies. Our Patient had a gross facial asymmetry along with loosening of 3rd molar tooth. No neurosensory deficits along the distribution of the trigeminal nerve were elicited.

In a study conducted by Becelli et al.[20] 60 patients were confirmed with mandibular ameloblastoma and it was found that about half of them showed typical symptoms such as swelling of the affected regions(38.3%),paresthesia of the innervated region of the mandibular nerve(13.3%) and alteration in dental occlusion in 10% of the cases.

Radiology and location are the key factors to diagnose ameloblastoma correctly. The diagnostic modalities can range from Lateral Oblique mandibular views ,Orthopantomographs, Computed Tomography (CT) scans and Magnetic Resonance Imaging (MRI). Radiographically the tumor appears as a multilocular cyst like lesion of the jaw with a classical soap bubble appearance. The lesion can also present itself as a unilocular variety. The periphery of the lesion is usually smooth on roentogram. In large and advanced lesions there might be thinning and expansion of the cortical plates. In our case CT scan imaging revealed characteristic large multilocular /soap bubble pattern of the lesion with thinning and expansion of cortices.

Bilkay et al. [27] in a retrospective analysis of 100 patients with benign mandibular lesion reported that a radiolucent lesion was found in 78% of the cases and 83% of them had cysts with well defined borders.

Ameloblastoma is known for its recurrence. The study of Henderson JM et al. suggests a local recurrence after initial ther-

apy. A recurrence rate of 50% to 72 % has been reported by them[28]. Recurrence may be attributed to method of treatment of the primary lesion, extent, site of origin.

Surgery is the treatment of choice for ameloblastomas. Treatment ranges from conservative approaches like curettage and enucleation to radical approaches by removal of some amount of normal bone beyond the tumor margins. Radiotherapy can also be an option but rarely used as primary line of treatment, it can be used for some inoperable cases.

In our case segmental resection of the left hemimandible was performed using the traditional approach for hemi mandibulectomy through a lip splitting and submandibular incision. The surgical access to remove a large tumor like our case could not be satisfied with other reported techniques in literature. Shirani et al[29] in a series of 7 patients have elaborated a new technique which is indicated for removal of large ameloblastomas of the mandible with immediate reconstruction only by using an intra oral incision. Its obvious advantage is maneuvering and repositioning of the mandible to remove the mass and to perform reconstruction simultaneously. This technique avoids facial scars and also bypasses the marginal mandibular nerve that innervates the lip.

Eppey et al. [30] in a comparative analysis reviewed 60 mandibular ameloblastoma cases and demonstrated that there was no recurrence in cases treated via en-bloc resection as compared to enucleation and curettage. The latter showed recurrence rate as high as 25% to 50%. Our case was a large ameloblastoma of the mandible with significant bone destruction visible on CT which required a more aggressive approach. A regular follow up of the patient has not revealed any signs of recurrence till date.

Surgical removal of large ameloblastomas leave large defects which are a challenge to repair. Mandible has to be reconstructed not only for aesthetics reasons especially in females but to improve overall functionality post surgery. A large untreated defect leads to severe midline shifts towards the operated sides leading to gross malocclusion. One of the biggest challenges in cases left without reconstruction is mastication and speech deficiency which have to be addressed immediately in the postoperative recovery phase to improve the quality of life of the patient.

There have been number of reconstruction procedures to treat large defects in literature. An ideal Treatment for large Ameloblastomas has been suggested by Chana et al. [31] in a series of 10 cases utilizing vascularized fibula flap and simultaneous placement of dental implants. Becelli et al.[27] describe two phases of reconstruction process in their study. The first phase comprises of reconstruction of the surgical defect by free or autogenous bone graft or revascularized autogenous bone graft. The second subsequent phase is of prosthetic rehabilitation by placement of dental implants. Another method of reconstruction

has been demonstrated by McCarthy et al. [32] by demonstrating internal distraction osteogenesis.

Cloke and Sandor[33] in a series of 10 patients with large mandibular defect have shown a latest technique of reconstruction by spanning the defects with rigid reconstruction plates to hold the remaining bone segments in position. The defects was stuffed with a bioimplant containing bone morphogenic protein-7(BMP-7) in a demineralized bone matrix(DBM)suspended in a reverse -phase medium to enable sustained BMP delivery.

In our case the resected defect was spanned with Titanium Reconstruction plates which provided a rigid support to both the lesser and the remaining mandibular segment. This provided our case with a descent acceptable aesthetic profile and functionality.

## CONCLUSION

Large ameloblastomas are always challenging to treat especially with conservative procedures like enucleation and curettage. Free wide marginal surgical excision of the tumor is the treatment of choice. This has both advantages and drawbacks. Radical approaches to treat large mandibular ameloblastomas on one hand reduce the recurrence rates but at the same time they create challenging tasks of reconstruction. A thorough assessment and algorithm of treatment plan has to be discussed along with Head & Neck ,maxillofacial and plastic surgeons to attain the best clinical outcome .

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