Case Report

Management of Giant Mandibular Ameloblastoma — A Case Report

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Ameloblastomas are rare, locally invasive and slowly growing and tumours with high recurrence rate. If they are not treated on time, they can reach an emormous size. Benign mandibular swellings are broadly divided into odontogenic and non-odontogenic tumours. Ameloblastoma is one of the most common benign tumours of odontogenic origin which developed from epithelial cells and its elements and dental tissues in their various phases of development. Patients with Giant Ameloblastomas are very rare but they are widely found and diagnosed in developing countries because of painless growth and patient's fear of Surgery leading to delayed treatment. This paper presents a case of large Ameloblastoma of left side of mandible in a 25-year-old female patient which was successfully resected in toto. Patient refused for free fibula flap re-construction of the defect because of her apprehension for donor site morbidity.

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Ameloblastomas or Adamantinomas are rare tumours of jaw it constitutes 1-3% of all jaw tumours[1]. It is more common in mandible, it presents as slow growing, painless swelling causing expansion and local destruction of cortical bone and they can grow to enormous size over the years without any malignant change. Hughes *et al* proposed that the term 'Giant Ameloblastoma' be reserved for lesions that are truly large, causes gross asymmetry and regional dysfunction. Treatment of such a lesion poses a challenge due to its extreme size, its dimensions and weight, extent and bones involved, vital structures involvement and the extent that it compromises the oral function[2]. The Giant Ameloblastomas are usually treated by Radical Surgery and it leaves a huge defect which demands re-construction[3]. A case of 25 years old female patient presented here with Giant Ameloblastoma was resected in toto.

Editor's Comment :

- Early detection and removal of such aggressive lesions of jaws can prevent major defects of the jaws which reduces functional as well as aesthetic quality of life.
- Aggressive Ameloblastomas need to be resected completely along with positive margins to prevent recurrence rate and second Surgeries.
- Radiographic and histopathological correlation is very important in such lesions. Long standing aggressive Ameloblastomas needs radiological and histopathological rule-out for further spread.
- Reconstruction of such lesion defects are important and necessary but patients decisions and affordability status needs to be respected too.

Case Report

A 25-year-old female patient presented with a history of a painless left mandibular swelling since a year which was progressively increasing in size. The swelling was associated with parasthesia of her left side of face mainly over the jaw and she also had difficulty in chewing due to the mass over the left mandible. There was a large firm swelling over the left mandibular region which was non tender extending from left ramus region extending the midline till lateral incisor of right lower quadrant. The Temporomandibular Joint (TMJ) movement was normal on both sides. The mouth opening was normal. The occlusion on left quadrant was completely deranged due to lesion expansion. Overlying skin was stretched and intact. The colour of overlying skin was normal.

The CT-SCAN of the lesion revealed a huge lytic lesion involving the mandible, the largest of its bulk was present around the left mandibular body and ramus. It crossed right mandible side and reached till the parasymphysis region. The lesion measured 116 x 109 x 134 mm (AP x TR x CC). Expanded mandibular cortex was partly thickened and partly invisible. This lesion had internal multilocular cystic and solid enhancing components. Multiple internal calcifications were seen within the lesion. A few of the involved teeth had resorption of their roots. Muscles at the floor of mouth were displaced with loss of fat planes where infiltration cannot be entirely ruled out.

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Posteriorly, it compressed left parotid, sub-mandibular glands & SCM muscle with resultant compression of left IJV. No evident infiltration. Mandibular condyle was not involved by the lesion.

Routine blood investigations were done and Fine Needle Aspiration Cytology (FNAC) incisional biopsy of the lesion was taken. FNAC from the swelling yielded a hemorrhagic, cystic fluid consisting of inflammatory cells, occasional epithelial cells, scattered RBCs and cholesterol crystals in a fibrinous background. The patient was provisionally diagnosed with multicystic Ameloblastoma. The surgery was planned under general anesthesia. Extended transcervical incision was made over the lesion and the tumour mass was exposed. The tumour was removed in toto keeping the safe margins of 1.5cm from the healthy bone. Hemimandibulectomy was done from lower right canine region distally and subcondyle region on the left side proximally. The condyle was preserved. Primary closure was done of the defect after achieving proper hemostasis. The patient was examined postoperative after 10-15 days. The healing is satisfactory without and relevant complications. Suture removal was done. The histopathology report of the lesion confirmed the lesion to be multicystic Ameloblastoma. The histological subtypes of such aggressive Ameloblastoma consisted of combination of follicular type and plexiform type of Ameloblastoma. The cells consisted of columnar or palisaded ameloblasts-like cells and triangular shaped cells seen in the inner zone similar to stellate reticulum. Also, the cells showed epithelium proliferating in a 'cord like fashion'; hence the histological type of ameloblastoma consisted of both follicular as well as plexiform type (Figs 1-7).

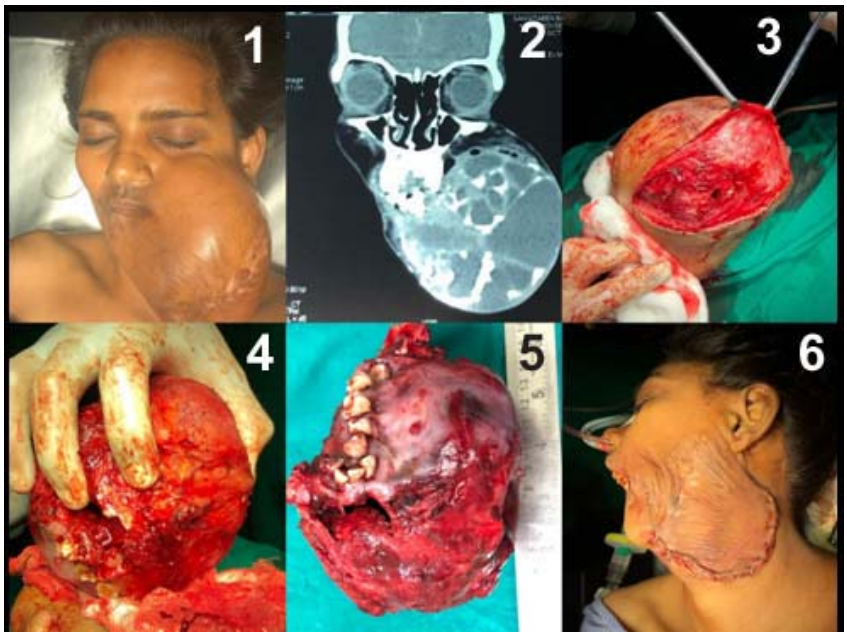

Fig 1 — Pre-operative view of the lesion; Fig 2 — Radiographic view of the lesion (CT-SCAN); Fig 3 — Cervical incision given for lesion exposure; Fig 4 — Lesion exposure and removal in toto; Fig 5 — Lesion removed completely; Fig 6 — Sutures taken after removal done

Discussion

Ameloblastoma are aggressive benign tumours originated from epithelium that may arise from the enamel organ, remnants of Odontogenic Keratocyst (OKC), dental lamina, dentigerous cyst or from the basal epithelial cells of the oral mucosa. It increases to great size and cause facial asymmetry, malocclusion, teeth displacement, loosening of teeth and pathologic fractures. Tumor size may range from 1 to 16 cm at presentation which result from bone expansion and soft tissue invasion. The clinical behaviour of the lesion is somewhere between benign and malignant lesion. The classification of the Ameloblastoma in the past was poorly deûned; the current concept is to classify the Ameloblastoma as solid/multicystic, classical

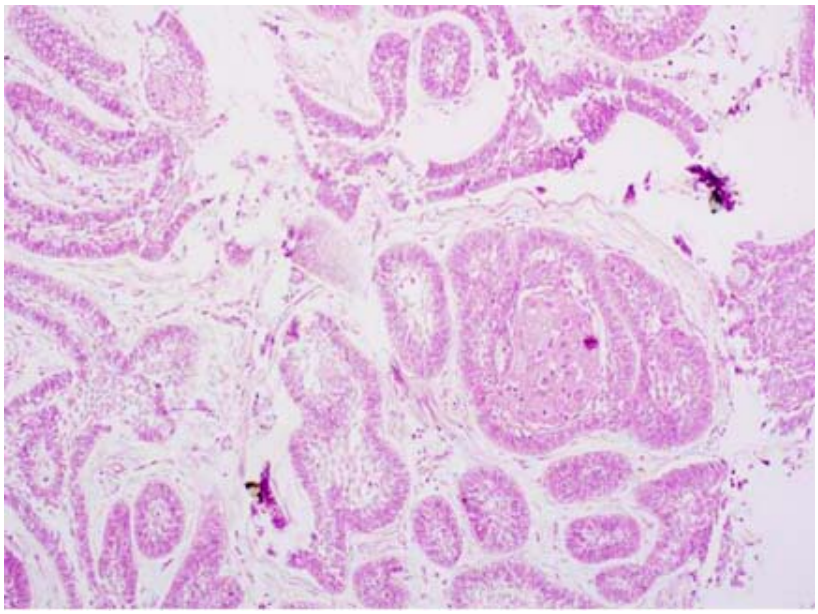

Fig 7 — Histopathological Report of the Lesion Confiming It to be Ameloblastoma (H&E, 10x)

intraosseous, peripheral or unicystic subtypes. This classification has direct bearing on the pathological behaviour of these variants. Solid or multicystic variants are aggressive locally and recur if they are not excised in toto. Unicystic Ameloblastoma was identified as distinct entity with less aggressive behaviour[4]. Treatment of ameloblastoma is primarily surgical. The conservative modalities include enucleation, cryosurgery and curettage and the radical modalities are marginal, segmental, and Hemiman-dibulectomy. The conservative modalities may be less aggressive but the recurrence rate up to 55–90% have been reported in the literature post it. Reconstruction of large mandibular defects as due to Giant Ameloblastomas poses a challenge. For mandibular and oral reconstruction, donor sites mainly include radial forearm, fibula, iliac crest and scapula. These sites are the primary sources of new vascularized bone and soft tissue. Among all these, fibula has many advantages including transfer of bone, soft tissue and skin[5].

Conclusion

Hence, the present case report gives a broad overview about a Giant Ameloblastoma of left side mandible which was resected completely. We recommend the radical approach for resection of these benign but such giant and locally aggressive tumours, resection with safe margin of at least 1.5-2 cm of healthy bone. Removal of lesion was done in toto with adequate safety margin and primary closure was done with fairly satisfactory cosmetic and functional results. Functional and esthetic results are better with free flaps, best among them being free fibula flap.

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