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Research Article

AN UNUSUAL LONG STANDING GIANT MANDIBULAR AMELOBLASTOMA RECONSTRUCTION USING NON-VASCULARISED ILIAC CREST: OUR EXPERIENCE

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ABSTRACT

Ameloblastoma of the jaws are true neoplasm of odontogenic epithelial origin, and derives its name from the English word "amel" which means enamel and the greek word "blastos" which means germ. It has an intermediate malignant potential, thus classified in a grey zone between benign and malignant neoplasms. It is the second most common odontogenic neoplasm with a incidence of approximately 1% of all oral tumors and 18% of all odontogenic tumors of jaws. More than 80% of cases of ameloblastoma occurs in the mandible. The ameloblastoma occurs mainly in three variants which are solid or multicystic, unicystic, and peripheral and painless expansion of the jaws with or without pain is the most common clinical presentation. Giant invasive ameloblastomas of mandible are rarely confronted. Although these ameloblastomas are benign, they are life threatening. Many have eventuated in death due to airway obstruction, starvation from restriction of feeding. We present a case of giant ameloblastoma in a 36 years old male which was a consequence of patient negligence. The lesion reached an enormous size over two decades (25 years) causing gross facial deformities and posed considerable problems in endotracheal intubation and resection.

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INTRODUCTION

Benign mandibular expansile lesions can be due to a wide variety of maxillofacial pathology which are divided into odontogenic and non-odontogenic lesions. Among these the most common odontogenic lesion includes, ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumor, central giant cell granuloma, fibro-osseous lesions and osteoma.[1] According to WHO 1992 ameloblastoma is defined as: Unicentric, non-functional, intermittent in growth anatomically benign, locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma.[2]

An ameloblastoma can occur at any anatomical location in both maxilla and mandible, but it is mandible that is involved in around 80% of cases.[3] Its peak incidence is in the 4th-5th decades of life and the male to female ratio is 1:1.[4] Neglected ameloblastomas may become enormous and cause gross facial deformities and upper airway obstruction that pose considerable problems in management. The different surgical treatment varies from curettage, enucleation to bony resection with or without reconstruction. Radiotherapy is not indicated as the lesion is radio-resistant. We present a case of approximately

14 x 13 cm giant mandibular ameloblastoma with 25 years of history & unbearable social problem. The lesion involves nearly whole mandible except the ramus and condyle of both sides, which was treated with resection with 0.5 cm safe margin and reconstruction using non-vascularised iliac crest graft.

Case Report

A 36-year-old male patient reported to the Unit of Oral and Maxillofacial Surgery with the complaint of swelling in the lower jaw since 25 years. The patient was operated for the lower jaw swelling during childhood, after that he noticed again swelling in lower chin region, the patient neglected the swelling owing to its painless nature and slow growth which gradually increased to present size. He also complains of difficulty in chewing & disfigurement of face along with respiratory distress in supine position, so he was always prefers lying in lateral recumbent position during sleeping. The swelling was associated with paraesthesia of lower lip on both side.

On physical examination, patient was moderately built and moderately nourished. On local examination, a large soft to firm swelling of mandible measuring approximately 14 x 13 cm (Fig.1). Prominent dilated veins were noticed on the surface of the lesion. Skin over the swelling appeared stretched.

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Fig 1 The patient presenting with giant mandibular swelling.

Intraorally a massive swelling was noticed in the floor of the mouth, pushing the tongue to the posteriorly and superiorly. Tongue movements were restricted with changed phonetics.

Computed tomography scan revealed a large expansile osteolytic lesion of mandibular origin, approximately measuring about 15 x 14 cm in size, showing multiloculated cystic and solid soft tissue components. Superiorly, it was extending up to bilateral angle of mandible and inferiorly up to the level of thyroid cartilage. The scan also showed buccolingual and antero-posterior expansion with endosteal scalloping, gross cortical thinning with areas of dehiscence and multiple cortical perforations. There was compromise in the nasopharyngeal airway space due to the lesion (Fig.2).

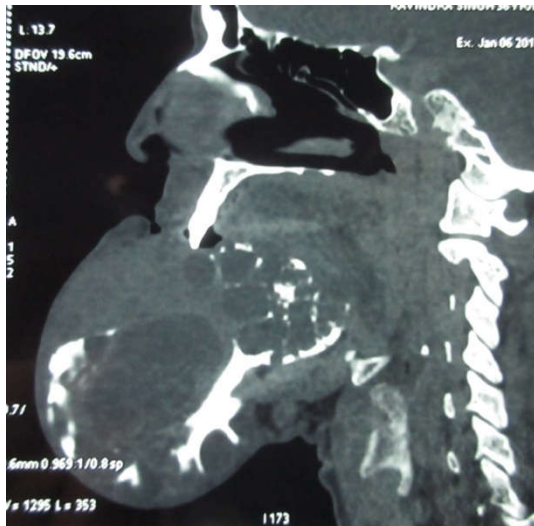


Fig 2 CT scan showed gross cortical expansion & compromised nasopharyngeal airway.

For diagnosis of this multilocular lesion, aspiration, followed by an incisional biopsy was performed under local anaesthesia. Needle aspiration was negative for blood or any clear fluids and, therefore, suggestive of a mass lesion. On biopsy, the tumor was diagnosed histologically as plexiform ameloblastoma.

The patient was taken under general anaesthesia for complete excision of the lesion. Extra oral incision was made directly over swelling by assuming incision 2 cm below lower border

(visor incision). Layer by layer dissection was done to expose the tumor. Approximately, 14 cm x 13 cm large solid tumor starting from left side of ramus to right side of angle crossing midline was noted (Fig.3).

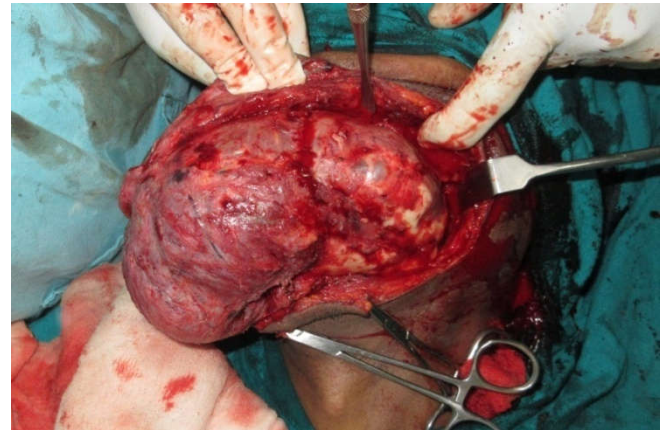


Fig 3 Intraoperative photograph after exposure of tumor.

Segmental resection was done and resected mandible shown in (Fig.4).



Fig 4 Resected mandible.



Fig 5 Six months post-operative photograph.

Reconstruction of the defect was done using anterior iliac crest graft and stabilized with reconstruction titanium plate. After removing extra tissue from stretched skin, regional muscles was sutured with recon plate on their assumed anatomical insertion and layer wise closure was done. The patient was followed up for 6 months with good post-operative symmetry and function (Fig.5).

DISCUSSION

The term extreme or giant odontogenic tumor like ameloblastoma should be reserved for the maxillofacial lesions that are truly huge causing gross facial asymmetry, respiratory distress and regional dysfunction.[5] The term ameloblastoma is coined by Churchill in 1933 but, the first detailed description of this lesion was by falkson in 1879.[6]

To some extent regional dysfunction like mild airway obstruction, masticatory and swallowing difficulties were observed in the present case. In recent times it is uncommon to find a large ameloblastoma involving near total of the mandible because of early detection owing to the easy access to the investigating aids and better awareness in society.

Ameloblastomas, though predominantly seen in the middle age group (4th – 5th decade of life), it is also known to occur in children (8.7% to 15%).[7] It may appear anywhere in the jaws, although approximately 80% of the lesion arise in the mandible, especially in the molar ramus region. Typically ameloblastoma present as painless slow growing mass as in our case, it took about 25 years before the patient reported with significant facial asymmetry, difficulty in chewing and deglutition. Around 13.3% of lesion presents with paraesthesia of the innervated region of mandible.[8]

The radiographic presentation of ameloblastoma is not pathognomonic and may vary from unilocular to multilocular radiolucency with well-defined margin and some time associated with impacted tooth. They characteristically exhibit slow but unrelenting and destructive growth. The multilocular radiolucency is found to be more common than the unilocular type.[9] It may expand the cortical plate which gives rise to a paper- thin and soap bubble appearance on panoramic X-ray as well as computed tomography scan.

Ameloblastoma is a tumour with a well-known propensity to recur due to its capacity to infiltrate trabecular bone. The solid variety has high propensity for local infiltration and therefore the highest potential for recurrence. It erodes the cortical bone and also spread in to the medullary space well beyond radiographic margins resulting in recurrence rate of 75-90% requiring more aggressive treatment.[10] Therefore the treatment of choice for such locally recurrent lesion is surgical excision with wide free margins, if not resection but, the growth characteristics of ameloblastoma vary from other benign lesions and tumour resection is often advocated regardless of the type of ameloblastoma. The appropriate amount of normal bone beyond the radiographic boundary required for a tumour-free margin has not been definitely established.

Some clinician suggest a resection of no less than 1-2 cm of normal appearing bone beyond the radiographic tumour margin.[9] Reconstruction of large defects of mandible had always been a challenge to the maxillofacial surgeons. The reconstruction aims in attaining acceptable cosmetic results and structural integrity that restores the functions like chewing, speech and swallowing to a better extent.

CONCLUSION

The challenge in the management of giant ameloblastoma of the jaw is not only to excise the tumour completely in order to prevent recurrence but also to provide the best reconstruction method for better facial appearance. Hence it is most emphasize, to both the clinician and the patient, the need for a definite treatment protocol and lifetime periodic follow-up for detection of recurrence as even a five year tumour-free period does not necessarily mean a cure. Satisfactory facial symmetry of the patient was restored without any recurrence. Hence, this surgical treatment for extensive mandibular ameloblastoma can obtain an excellent result by the shortest time and the lowest economical cost.

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