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CASE REPORT

Single Stage Management of Giant Ameloblastoma

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ABSTRACT

Ameloblastomas are tumors of odontogenic epithelium. Although benign, they are quite aggressive, invasive, and at times reach gigantic proportions. Patients with giant ameloblastomas are very rare, but are still encountered in developing countries due to painless growth and patient's fear of surgery leading to delayed treatment. Management of giant ameloblastomas poses a challenge in terms of acceptable functional and esthetic outcome. It has been advocated that large ameloblastomas are best managed in single stage procedures which involves resection and reconstruction, thus reducing patient stay, cost factor and related morbidity. This paper presents a case of large ameloblastoma of the mandible of 25-year-old male which was successfully managed in a modified single stage procedure, thus restoring patient function and esthetics.

Keywords: Giant ameloblastoma, Mandible, Single stage treatment.

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INTRODUCTION

Ameloblastomas are benign, epithelial, odontogenic, jaw tumors constituting about 1% of all oral tumors and 9 to 11% of odontogenic tumors with an incidence rate of three per 10 million.[1] It is a tumor characterized by local aggressiveness, multiple recurrences and a metastatic potential. Hughes et al[2] 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 that involves questions pertaining to its dimensions and weight, the number of bones involved, extension to vital structures, and the extent of compromise in oral functions.[2] The aggressive lesions are usually treated by radical surgery that frequently leaves behind large jaw defects.[3] Few cases of large ameloblastomas have been reported that were surgically excised followed by immediate reconstruction using microvascular grafts from scapula, iliac crest, or fibula.[4-7] A case of 25-year-old male patient is presented that was resected and reconstructed with fibula microvascular graft in a single operative procedure. The esthetic and functional outcomes are extremely satisfactory. This case highlights that single stage management is better with vascularized bone graft after radical resection of the lesion. Thus, this case highlights the benefits of single stage radical management of giant ameloblastoma over conservative or multistage procedures.

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CASE REPORT

A 25-year-old male patient visited our department of oral and maxillofacial surgery, King George's Medical University, Lucknow with chief complaint of progressively slow growing, painless, nontender swelling over left side of face for last 12 years. Patient's history revealed of slowly progressive lesion with a sudden onset. Patient did not undergo any surgical procedure previously.

On extraoral inspection, the face is asymmetrical from frontal view (Fig. 1) with lesion extending vertically from zygomatic arch to lower border of mandible and horizontally from corner of the mouth to left ear lobe. The ear lobe is not lifted. The lesion is spherical in shape measuring 14 × 14 cm in both vertical and horizontal dimensions. The surface of the lesion is smooth with well-defined edge and tensed overlying skin. On palpation, there is no local rise in temperature, variable consistency, presence of fluctuations and fluid thrill in certain regions. The lesion is nonreducible and noncompressible without any fixity to overlying skin. The left submandibular lymph nodes are palpable, tender with 5 × 5 mm in dimensions. There was no anesthesia or paresthesia reported in the distribution of inferior alveolar or mental nerve. On intraoral inspectory findings, the lesion extends from first premolar of left side to ramus with expansion of lingual cortical plate. The overlying mucosa is normal in color, texture, and consistency without the presence of any draining sinus (Fig. 2). Radiographic examination with computed tomography (CT) scan revealed the presence of well-circumscribed multilocular lesion involving the left side of the mandibular body to involve the neck of condylar

process. There are areas of lingual perforation of the cortical plate in certain areas (Fig. 3). Normal blood investigations were done and patient was informed about the disease and written consent was taken for fine needle aspiration cytology (FNAC) and incisional biopsy of the lesion. 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. Incisional biopsy of the lesion reveals findings, such as proliferation of tall columnar (ameloblast like) cells in the form of anastomosing cords with central stellate reticulum like cells with underlying dense connective tissue stroma (Fig. 4). Following a definitive diagnosis of multicystic ameloblastoma chest radiograph and CT scan abdomen was done to rule out metastasis. Surgery was planned under general anesthesia. An extended submandibular incision was given to expose the tumor mass. The tumor was removed keeping a safe margin of 1.5 cm of sound bone in distal segment whereas disarticulation was done in proximal segment (Fig. 5). The left lower limb was prepared for harvesting of vascularized fibula graft. A sigmoid incision was given on lateral side of lower left leg in the region of intermuscular septum. Dissection was done by raising thigh tourniquet at a pressure of 350 mm Hg. Dissection is continued anteriorly of fibula and peroneus longus, peroneus brevis, and extensor hallucis longus muscles are elevated. After the fibula is distracted, peroneal artery and vein can be seen along the medial aspect of fibula. This is transacted along with flexor hallucis muscle. The fibula along with the vessels is harvested preserving the periosteum and muscles (Fig. 6). Microsurgical anastomosis is done between peroneal artery to facial artery and peroneal vein to facial vein (Fig. 7). Layer-wise closure was done with 3-O vicryl in muscular and subcutaneous tissues and 4-O nylon in skin. The sample was sent for histopathological exami-

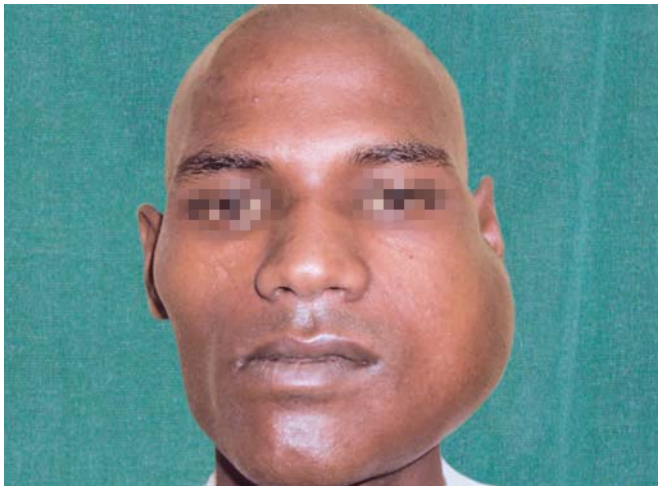

Fig. 1: Asymmetrical face showing enlargement of mandible over left side of face

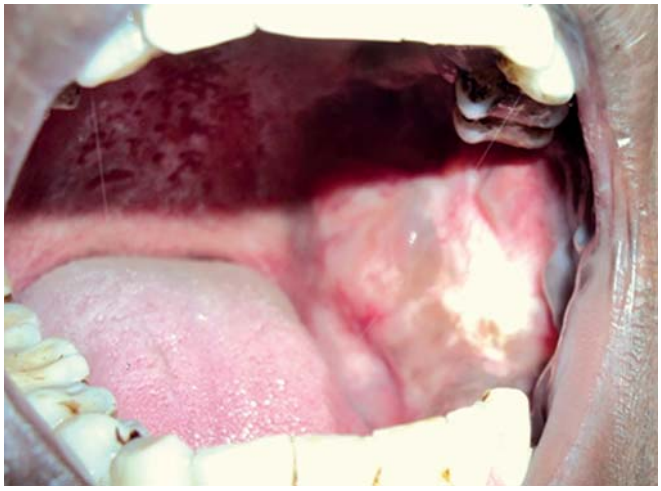

Fig. 2: Intraoral picture of the lesion showing normal overlying mucosa in texture and consistency

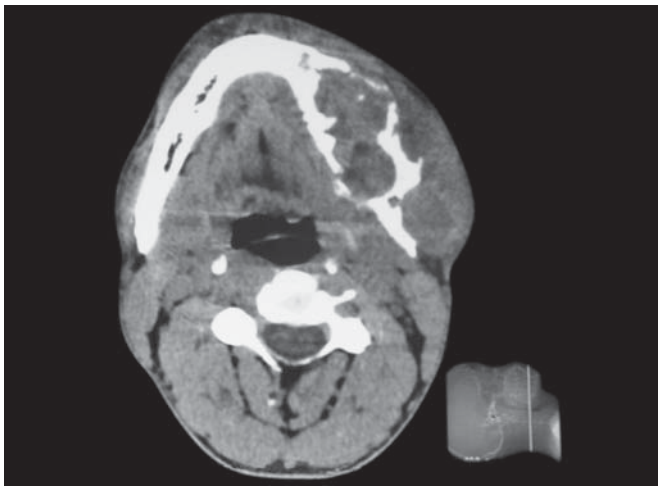

Fig. 3: Axial view of the CT scan revealing both buccal and lingual perforation of cortical plate

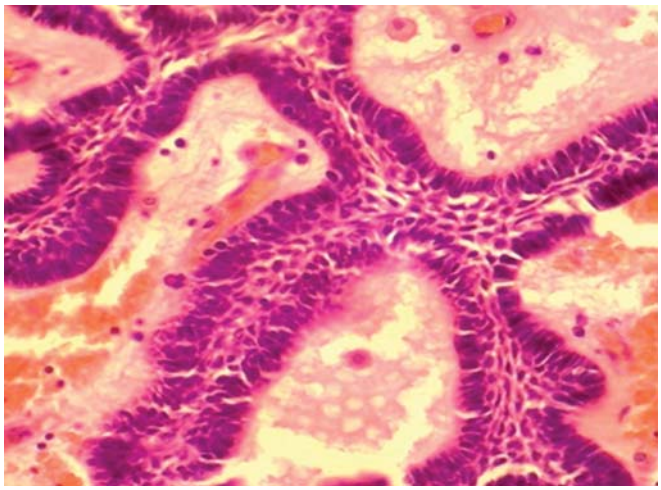

Fig. 4: Histopathological picture depicting proliferation of tall columnar (ameloblast like) cells in the form of anastomosing cords with central stellate reticulum like cells with underlying dense connective tissue stroma suggestive of ameloblastoma

nation for reconfirmation of previous diagnosis. The patient recovered uneventfully with desirable esthetic and functional outcome when followed for a period of 1 year both clinically (Fig. 8) and radiographically in OPG (Fig. 9). There is no motor defect in lower limb with normal dorsiflexion of great toe due to associated injury to peroneal nerve.

DISCUSSION

Ameloblastoma with their aggressive clinical behavior and benign histopathological features as such constitute a puzzling paradox. Ameloblastoma typically presents as a painless slow growing mass and can cause facial asymmetry, displacement of teeth, pathological fractures to name a few. Apart from facial asymmetry, other symptoms due to ameloblastoma include difficulty in mastication, malocclusion, pain and paresthesia of the affected area.[1] Neglected ameloblastomas become enormous and can cause gross facial deformity, oral dysfunction and considerable management problems.[2] Since the introduction of panoramic radiography in routine dental practice, it has been rare to find large ameloblastomas involving half of the mandible.[3] Giant ameloblastomas are rare in developed societies but can occur in patients who delay treatment because of fear of surgery. In the present case, patient simply did not report due to lack of any oral and regional dysfunction and also due to phobia of surgery. Giant ameloblastomas may be associated with secondary hypoproteinemia, due to protein loss into cystic spaces of the tumor which sometimes form fistulous tract.[8] The poor nourishment attributed to the tumor also lead to malnourishment of the patient which lead to anemia. Hence, preoperatively hematinic syrups were administered to improve the condition of the patients. The treatment modality of giant ameloblastomas includes surgical excision of tumor with wide free margins followed by reconstructive procedures using autogenous bone graft. With the advent of vascularized osseous free flaps over the last three decades, it is the preferred reconstructive modality currently for mandibular defects, with success rates of over 90%.[4-7] Reconstruction of large mandibular defects as due to giant ameloblastomas poses a challenge to the head and reconstructive surgeons. Reconstruction may either be done as part of the initial surgery as one stage treatment or can be deferred for second stage, few weeks after the primary surgery. Besides reducing the inpatient stay and cost of treatment, single stage procedure also reduces the number of surgeries and the associated morbidity and mortality rate, while restoring normal esthetics and function.[9] The rate of recurrence and chances of distant metastasis are also low. The mandible is both cosmetically and functionally an important structure of head and neck, contributing to facial appearance, speech, mastication and swallowing.[6] For mandibular and oral reconstruction, four donor sites, i.e. fibula, iliac crest, radial forearm, and scapula have become the primary sources of vascularized bone and soft tissue. Among all these, fibula has multiple advantages including bone length and thickness, donor site location permitting flap harvest simultaneously

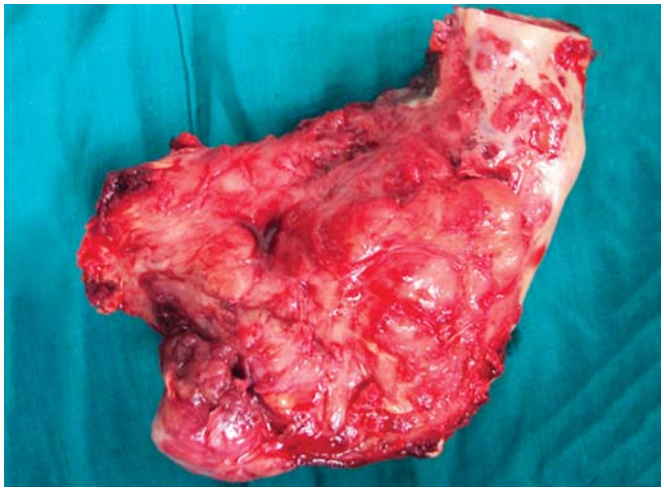

Fig. 5: Resected specimen disarticulated from TMJ

Fig. 6: Harvested fibula along with feeding vessels

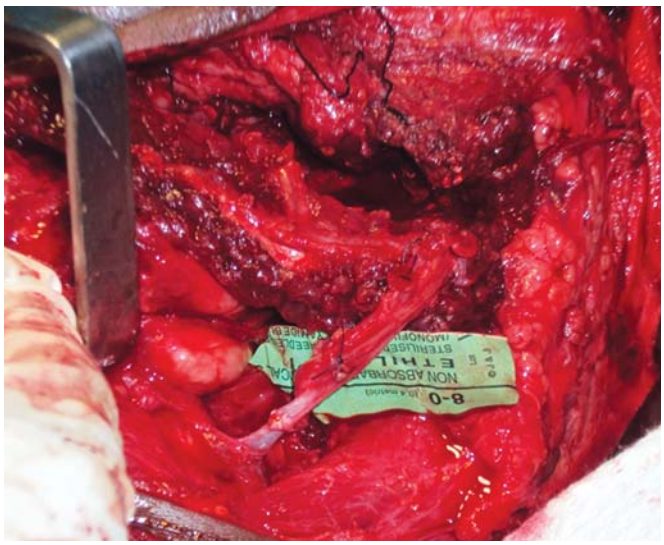

Fig. 7: Microsurgical anastomosis is done between peroneal artery to facial artery and peroneal vein to facial vein

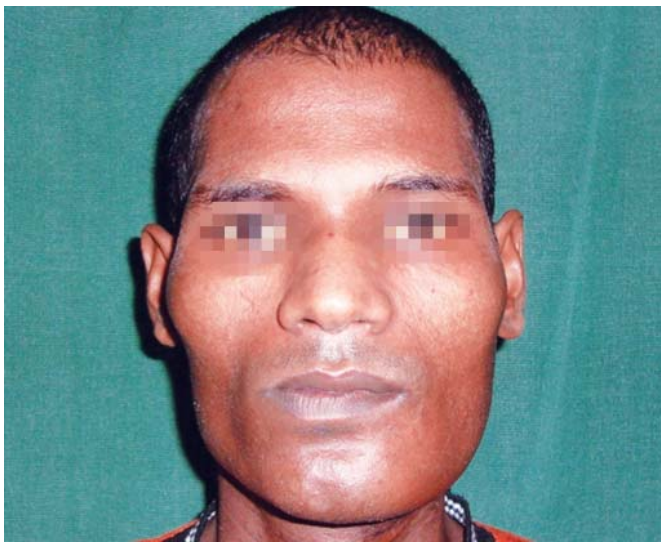

Fig. 8: Final outcome of the patient, 1 year postoperatively

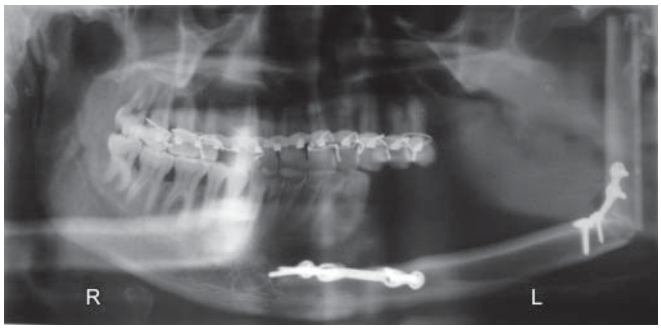

Fig. 9: Postoperative OPG of the patient showing good uptake of the fibula graft after 1 year postsurgical period

with tumor resection because both teams are at different end of the table, has least resorption and failure rate, minimal donor site morbidity and therefore should be considered as the first choice.[4-7] In fact, few retrospective case studies have shown that giant ameloblastomas treated in single procedure by resection and reconstruction with fibula free flap has had a successful outcome.[10,11] Gerzenshtein et al[10] in a follow-up of 2 to 3 years for three cases of giant ameloblastomas treated with fibula flap concluded that it is a reliable reconstructive operation for giant ameloblastomas. Chaine et al[11] in follow-up periods spanning up to 7 years for nine cases of giant mandibular ameloblastomas, also affirmed that free fibular flap reconstruction was an effective treatment modality for such cases. Recently, other reconstruction methods like distraction osteogenesis and bioimplant containing BMP-7 have been tried but have some limitations like very long treatment span.[6]

Hence, the present case report gives a broad overview about a giant ameloblastoma and a single stage management with vascularized bone grafts which benefits the patient from large mandibular defects.

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