Case Report

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Follicular variant of ameloblastoma mandible reconstructed with free fibula: a case report

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ABSTRACT

Ameloblastoma is a tumor of odontogenic epithelium arising from remnants of dental lamina. It is a benign tumor but locally invasive affecting mandible more than maxilla. Ameloblastomas grow into the bone and soft tissues causing facial disfigurement and facial deformities. Treatment is surgical. We present a case report of 60 year old male with swelling in jaw, biopsy revealed ameloblastoma. Anterior segmental mandibulectomy was done and reconstructed with fibular flap which restored both function and aesthetic appearance.

Keywords: Ameloblastoma, Mandible, Multicystic, Reconstruction

INTRODUCTION

Ameloblastoma is a benign tumor of odontogenic epithelium principally of enamel organ-type tissue.1 It accounts for about 1% of all oral tumors and about 9-11% of odontogenic tumors.² It is a benign tumor but locally aggressive. Ameloblastoma accounts for 60.3% of all odontogenic tumors in Indian population.³ There is a male preponderance and majority ameloblastomas occur in mandibular ramus region. Although ameloblastomas are primarily intraosseus tumors, they have been occasionally reported in soft tissues. They are classified into unicystic, multicystic peripheral and desmoplastic (solid), types. Ameloblastoma in the mandible can progress to large size and cause facial asymmetry, displacement of teeth, malocclusion, and pathologic fractures.

CASE REPORT

We present a case of 60 year old male with primary complaints of swelling in his left jaw from the last 10 years. It was insidious in onset and gradually progressive

and associated with malocclusion of teeth (Figure 1). There was no swelling in the neck and there were no other ear and nasal complaints. Examination revealed 8x4x4 cm hard swelling on the left side of mandible involving the ramus and body.



Figure 1: Preoperative image showing a left sided lesion involving body of mandible.

The swelling was non tender and free from overlying skin. On oral examination molar teeth malocclusion was present and there was expansion of alveolus with extension to floor of mouth. There was no neck lymphadenopathy and rest of ENT examination was normal. Contrast enhanced computed tomography of the mandible was done which showed 65 x 32 mm well defined expansile multiseptated lytic lesion in symphysis menti extending to body of mandible with thinning of inner and outer cortex making a differential diagnosis of ameloblastoma (Figure 2). Fine needle aspiration cytology of swelling was done which was reported as ameloblastoma.

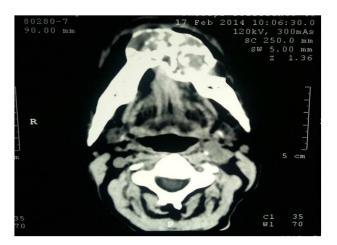


Figure 2: Contrast enhanced CT showing mandibular lesion with erosion of inner and outer cortex.

Patient underwent segmental mandibulectomy with a margin of one cm of normal mandible. Segmental mandibulectomy was done. Neck skin crease incision was made about two finger breadth below the mandible, sub platsymal flap was elevated and swelling was identified. Osteotomies were done on both sides and tumor was removed (Figure 3 and Figure 4). The resultant defect was reconstructed with free fibular flap harvested from right leg.



Figure 3: Surgical exposure of lesion and osteotomy cut made with 1.5 cm of normal mandible.



Figure 4: Resected specimen.

Arterial anastomosis was made with facial artery and venous anastomosis was made with external jugular vein. Fibula was carved into mandible by osteotomies and fixed to remaining mandible with help of miniplates. Hyomandibulopexy was done by drilling holes in body of hyoid bone and through the fibular flap and passing a 1-0 prolene suture (Figure 5).

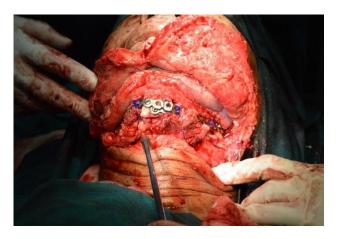


Figure 5: Free fibula carved into mandible fixed with miniplates and hyomandibulopexy being performed.

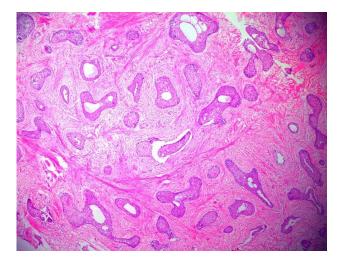


Figure 6: Histopathological image showing the follicular variant of ameloblastoma.

This helps in elevating the hyoid with mandible so crucial for initiation of swallowing. Skin paddle of the flap was sutured with the floor of mouth. The specimen was sent for histopathological analysis which was reported as follicular variant of solid ameloblastoma (Figure 6), all the margins were free. Patient received feeding through ryle's tube for the first seven days and later oral feeding was started. Patient was followed every month for first three months and then six monthly thereafter. The patient experienced a favorable recovery without any donor or recipient site complications, and a favorable aesthetic and functional outcome was observed during the 12-month follow-up period.

DISCUSSION

Ever since the term "Adamantinoma" was coined by Mallassez, controversy has prevailed as to the most appropriate form of treatment to prevent recurrence of the lesion. The term ameloblastoma was coined by Ivey and Churchill.⁴ Ameloblastomas account for 1% of benign tumors and cysts of the jaw.² Incidence is estimated to be 0.5 cases per million person-years worldwide.²

The study by Eversole et al, ameloblastomas were classified according to the histological findings into follicular, plexiform, acanthomatous, granular cell, basal cell, squamous metaplastic, and other rare types. Ameloblastomas are also classified into unicystic, multicystic, peripheral and desmoplastic types. Robinson on reviewing 293 cases reported site incidence of 83.7% in the mandible and 16.3% in the maxilla. Multicystic/solid is the most common form of ameloblastoma. Ameloblastomas can present with huge swellings over the jaws which can result in disturbances in facial aesthetics and function, such as difficulty with mouth opening, swallowing, chewing, breathing, neurologic deficits, and pathologic fractures.

Multicystic ameloblastoma usually present in the fourth-fifth decade and there is slight male gender predilection. Majority of them occur in the posterior region of mandible.

The lesions more often progresses slowly and are locally invasive and infiltrates through the medullary spaces and erodes cortical bone. If left untreated, they resorb the cortical plate and extend into adjacent tissue. Crepitation or eggshell crackling may be elicited. Posterior maxillary tumors might obliterate the maxillary sinus and may extend intracranially and adjacent tissues like orbit imposing a serious challenge for excision. Unlike mandibular tumors, maxillary tumors often go undetected in early stages.

Radiographically solid/multicystic ameloblastoma show an expansile, radiolucent, multiloculated cystic lesion, with a characteristic "soap bubble-like" appearance. Other findings include cystic areas of low attenuation with scattered regions representing soft tissue components. There can be thinning and expansion of the cortical plate with erosion with the displacement and resorption of adjacent teeth.⁸

Several treatment modalities have been tried by various authors which include Curettage, Enucleation, Chemical cauterization, Electro cauterization, En bloc excision, Radical resection and Radiotherapy/chemotherapy. Sehdev et al. in 1974 analyzed 72 patients with ameloblastoma of mandible and they concluded that the conservative approach (curettage) led to 90% recurrence of mandibular ameloblastoma.9 Shatkin in 1965 reported twenty cases of ameloblastoma and observed that 86% of the mandibular lesions recurred after curettage compared to a 14% recurrence rate after en bloc resection.¹⁰ Atkinson et al. reported the use of mega voltage radiation for ten patients with recurrent ameloblastoma.11 Five of the six patients with mandibular tumor had a reduction in tumor size only. There is a danger of postradiation sarcoma as reported by Becker and Pertl in 1967 and of osteoradionecrosis.12

Muller and Slootweg and Curi defined radical treatment as a definitive procedure in which the intention was to remove the ameloblastoma with a margin of about one cm of normal bone. 13,14 Radical wide resection with safety margins and subsequent reconstruction is generally recommended. A fibular free flap (FFF) is commonly used to reconstruct the mandible in order to adequately restore both aesthetic appearance and function. In the past, mandibular defects presented a major challenge because of the limited reconstruction options with a pedicled flap. However, current advances in microsurgery have allowed for composite soft and bony facial reconstruction. Fibula, scapula, and iliac crest flaps are commonly used as donor sites to reconstruct composite mandibular defects after ablative surgery.

The main modality of treatment is surgery, with wide resection recommended due to the high recurrence rate of solid/multicystic ameloblastomas. The recurrence rate after resection is 13-15%, as opposed to 90-100% after curettage so a margin of 1.5-2 cm is recommended beyond the radiological limit to ensure that all microcysts are removed.¹⁵

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REFERENCES

- Nakamura N, Mitsuyasu T, Higuchi Y, Sandra F, Ohishi M. Growth characteristics of ameloblastoma involving the inferior alveolar nerve: A clinical and histopathologic study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;91:557-62.
- 2. Becelli R, Carboni A, Cerulli G, Perugini M, Iannetti G. Mandibular ameloblastoma: Analysis of

- surgical treatment carried out in 60 patients between 1977 and 1998. J Craniofac Surg. 2002;13:395-400.
- 3. Krishnapillai R, Angadi PV. A clinical, radiographic, and histologic review of 73 cases of ameloblastoma in an Indian population. Quintessence Int. 2010;41:e90-100.
- 4. Ivey RH, Churchill HR. The need of a standardized surgical and pathological classification of tumors and anomalies of dental origin. Am Assoc Dent Sch Trans. 1930;7:240-5.
- Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. J Oral Maxillofac Surg. 1984;42:735-40.
- 6. Robinson HB. Ameloblastoma: A survey of three hundred and seventy nine cases from the literature. Arch Pathol. 1937;23:831-43.
- 7. Mendenhall WM, Werning JW, Fernandes R, Malyapa RS, Mendenhall NP. Ameloblastoma. Am J Clin Oncol. 2007;30:645-8.
- 8. Dunfee BL, Sakai O, Pistey R, Gohel A. Radiologic and pathologic characteristics of benign and malignant lesions of the mandible. Radiographics. 2006;26:1751-68.
- 9. Sehdev MK, Huvos AG, Strong EW, Gerold FP, Willis GW. Proceedings: Ameloblastoma of maxilla and mandible. Cancer. 1974;33:324-33.
- 10. Shatkin S, Hoffmeister FS. Ameloblastoma: a rational approach to therapy. Oral Surg Oral Med Oral Pathol. 1965;20(4):421-35.

- 11. Atkinson CH, Harwood AR, Cummings BJ. Ameloblastoma of the jaw. A reappraisal of the role of megavoltage irradiation. Cancer. 1984;53:869-73.
- 12. Becker R, Pertl A. On the therapy of ameloblastoma. Dtsch Zahn Mund Kieferheilkd Zentralbl Gesamte. 1967;49:423-36.
- 13. Müller H, Slootweg PJ. The growth characteristics of multilocular ameloblastomas. A histological investigation with some inferences with regard to operative procedures. J Maxillofac Surg. 1985;13:224-30.
- 14. MM, Dib LL, Pinto DS. Management of solid ameloblastoma of the jaws with liquid nitrogen spray cryosurgery. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1997;84:339-44.
- 15. Hong J, Yun PY, Chung IH, Myoung H, Suh JD, Seo BM, et al. Long-term follow up on recurrence of 305 ameloblastoma cases. Int J Oral Maxillofac Surg. 2007;36:283-8.

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