Anterior mandibular ameloblastoma

Ajay H. Bhandarwar,1 Girish D. Bakhshi,1 Ashok D. Boris,1 Amol Wagh,1 Rajat Kapoor,2 Channabasappa G. Kori1
1Department of Surgery and 2Department of Plastic & Reconstructive Surgery, Grant Medical College & Sir JJ Group of Hospitals, India

Abstract

Ameloblastoma is a benign odontogenic tumor. These are usually asymptomatic until a large size is attained. Ameloblastoma has tendency to spread locally and has a high recurrence rate. Majority of ameloblastomas (80%) arise from the mandible. Ameloblastoma arising from anterior mandibular region (symphysis-menti) is rare. Very few cases of midline anterior ameloblastomas are reported in the literature. They often require wide local excision. Reconstruction of mandible in these cases is challenging. We present a case of mandibular ameloblastoma arising from symphysis-menti. Patient underwent wide surgical excision of the tumor followed by immediate reconstruction using free fibular vascular flap, stabilized with titanium reconstructive plates. A brief case report and review of literature is presented.

Introduction

Ameloblastoma (Adamantinoma) is a benign odontogenic epithelial tumor.1 It accounts for 1% of all endodermal tumors and 11% of odontogenic tumors. It arises most frequently from the residue of dental ledge, enamel organ or Malassez epithelial rests or basal layer of oral epithelium.2,4 The exact aetiology is not known.2 The tumor frequently develops in the mandible (80%), maxilla (16%) while peripheral adamantinoma located in the soft tissue account for remaining 4%.2 In the mandible, it frequently involves molar & mandibular angle (70%), premolar (20%) and rarely anterior region (10%).2 It is a slow growing tumor, with equal gender prevalence. These are usually asymptomatic and presents with bony deformity.

Preoperative imaging including orthopantomogram (OPG), computed tomography (CT scan), magnetic resonance imaging (MRI) and 3D CT scan play important role in planning extent of surgical resection. Treatment modalities include surgical excision, enucleation, curettage, cryotherapy, radiotherapy and chemotherapy. Wide surgical excision with safe margins is the preferred treatment method.

We present a rare case of anterior mandibular ameloblastoma (<10%) arising from symphysis-menti, for which segmental resection of mandible was done with simultaneous reconstruction using free fibula vascular flap stabilized with reconstructive titanium plates.

Case Report

A 42-year-old male patient was admitted with lower jaw swelling of three years duration. Initially he was asymptomatic, however he then complained of bony deformity with gradual spontaneous fall of lower central teeth, difficulty in opening mouth and chewing food since 3-4 months.

On examination, patient had a huge bony tumor of the mandible with a parchment crunch feel. It was extending from left second premolar to the first premolar on right, predominantly affecting anterior segment of mandible. Overlying skin was freely mobile (Figure 1a). Intraorally, tumor was extending into sublingual region displacing tongue with loss of lower incisor and canine teeth (Figure 1b). There were few mucosal ulcerations. No cervical lymphadenopathy was noted.

OPG displayed a multilocular, osteolytic defect predominantly around midline extending to left and right causing marked expansion in the lower part with thinning of cortex of the mandible. A 3D CT scan revealed the presence of an expansive, multilocular, lytic lesion centered on the symphysis-menti (Figure 2a) and extending into parasympyseal region and into the proximal body of the mandible on the left side (Figures 2b and 2c). It was extending exophytically both anteriorly and inferiorly measuring 7.4x4.7x4.7 cm in size.

Wide local excision of central part of mandible was performed with simultaneous reconstruction using free fibular vascular flap, stabilized with titanium plates.

Correspondence: Ajay. H. Bhandarwar, 702, Vibhavari, Dinshaw wacha road, Churchgate, Mumbai-400020, Maharashtra, India. Tel. +91.0.9869039609 - Fax: +91.22.23735599. E-mail: abhandarwar@gmail.com

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Figure 1. Preoperative external appearance of lower jaw swelling. (a) Frontal view. (b) Intraoral view showing tumor extending into sublingual region.

Figure 2. 3D computed tomography revealing large expansile, multiseptate, lytic lesion involving (a) anterior portion of mandible. (b, c) tumor extension into left lateral part of the mandible.
stabilized with a titanium reconstructive plates (Figure 3).

Gross specimen showed few cystic areas with more solid component (Figure 4a) and cut section revealed cystic areas with few areas of necrosis and hemorrhage (Figure 4b).

Histology confirmed ameloblastoma with negative free margin (Figure 5a). Immunohistochemistry report revealed ameloblastoma positive for cytokeratin 5 and 6 (Figure 5b) and cytokeratin 14, negative for CD 68, cytokeratin 8 and 18 and calretinin.

Postoperative course was uneventful. Follow up x-rays showed almost normal contour of lower jaw (Figure 6a) and good cosmesis (Figure 6b) with normal jaw movements and normal healing bone graft. Patient is on follow up for prosthetic rehabilitation to restore missing tooth (dental implantation).

Present case highlights the importance of functional reconstruction along with cosmesis in an adult male after wide excision of anterior mandible.

Discussion

Ameloblastoma (from the early English word *amel*, meaning enamel and the greek word *blastos*, meaning germ) is a rare, benign odontogenic tumor of epithelial origin. It was described in 1827 by Cusack and designated as an Adamantinoma in 1885 by the French physician Louis-Charles Malassez and renamed as Ameloblastoma in 1930 by Ivey and Churchill.1 Unicystic ameloblastoma, a variant was first described by Robinson and Martinez.2

Ameloblastoma are usually diagnosed between the 4th and 5th decades of life except in unicystic variety (accounts for 6%), which is diagnosed between the ages of 20 and 30 years. No gender predominance is noted.5 About 10-15% of the tumors are often linked to a nonerupted tooth (impacted wisdom tooth).7 Ameloblastoma are usually asymptomatic and found on routine dental x-rays; however they present with jaw expansion. Its slow but relentless growth may cause movement of tooth roots or root resorption. In our case patient had lower jaw swelling with bony deformity and gradual spontaneous fall of lower central incisor and canine teeth, difficulty in opening mouth and chewing food.

Radiographically, it can either be unicystic intraosseous, multicystic, solid intraosseous (80-90%) or peripheral.5 These are histologically classified into follicular, granular, lexiiform, desmoplastic, basal cell and acanthomatous variety.8 Diagnosis is usually made by fine needle aspiration, which reveals palisaded basal cell layer with stellate reticulum like epithelium. Immunohistochemistry study revealed tumor cells positive for keratin and express CK 5, 6 and 14, few cells also reactive for cytokeratin 8, 18 and 19. Calretinin is expressed by certain solid and multicystic ameloblastoma. Granular cell ameloblastoma specifically express CD 56 and S-100 protein along with keratins. In the present case, ameloblastoma was of follicular variety and on immunohistochemistry study tumor cells, positive for cytokeratins 5 and 6 and cytokeratin 14, negative for CD 68, cytokeratin 8 and 18 and calretinin.

According to World Health Organization histological typing of odontogenic tumor, second edition 1992, ameloblastoma are further subclassified as summarized in Table 1.10

| Benign | Malignant |
|--------|-----------|
| Odontogenic epithelium without odontogenic ectomesenchyme | Odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation |
| i. Ameloblastoma; | i. Ameloblastoma fibroma; |
| ii. Squamous odontogenic tumor; | ii. Ameloblastoma fibrodeninima and fibro-odontoma; |
| iii. Clear cell odontogenic tumor; | iii. Odontoameloblastoma; |
| iv. Calcified epithelial odontogenic tumor. | iv. Adenomatoid odontogenic tumor; |
| v. Complex/ compound odontoma; | |
| vi. Calcified odontogenic cyst. | |

Table 1. Ameloblastoma subclassification, according to World Health Organization (1992).
transformation of pre-existing ameloblastoma. These usually occur in adults, aggressive in nature with poor outcome. Most common site of spread includes lung, cervical lymph nodes and rarely brain.

Surgery is the mainstay of treatment and involves complete removal of tumor with negative margin of 15-20 mm. Segmental resection or complete enucleation of mandible with simultaneous and final reconstruction of the postoperative defect gives normal contour and better functional ability which is only possible when the patient’s general condition is good. A free vascularized fibula flap offers the best option for mandibular reconstruction and subsequent dental implantation. The autogenic bone transplant from the iliac ala, fibula, scapula or radius is currently believed to be one of the best methods for mandible reconstruction. In most cases described in literature the results are satisfactory both in terms of functionality and aesthetics. There is little information available in the literature regarding the role of radiotherapy, as a useful modality of treatment in case ameloblastoma. Role of chemotherapy is not yet well defined, either single agent or combination chemotherapy have been used for palliation in patients with irresectable metastasis but only few reports showed little response (Cisplatin and Paclitaxel). Among surgical modalities, recurrence rates were 3.6% for wide resection, 30.5% for enucleation and 16% for enucleation followed by use of Carnoy’s solution. Few reports are available in the literature regarding recurrence of ameloblastoma in the grafted bone. Hence long-term follow up is recommended for more than 10 years at regular intervals after appropriate surgical operation. Patient should be on clinical and radiological follow up irrespective of the treatment done.

References

1. Malassez L. Sur le role des debris epitheli-aux papdentaires. Arch Physiol Norm Pathol 1885:6:379-449.
2. Niedzielska I, Pajak J, Langowska-Adamczyk H. Szkliwiak Szczeki - opis dwuch przypadków I przegląd pismiennictwa. Czas Stom 2004:57:255-60.
3. Martins WD, Favaro DM. Recurrence of an ameloblastoma in an autogenous iliac bone graft. Oral Surg Oral Pathol Oral Med Oral Radiol Endod 2004:98:657-9.
4. Al-Khaleeb T, Ababneh KT. Ameloblastoma in young Jordanians: a review of clinicopathologic features and treatment of 10 cases. J Oral Maxillofac Surg 2003;61:13-8.
5. Robinson L, Martinez MG. Unicystic ameloblastoma: a prognostic distinct entity. Cancer 1977;40:2278-85.
6. Stanley HR, Diehl DL. Ameloblastoma potential of follicular cyst. Oral Surg Oral Med Oral Path 1965;20:260-8.
7. Arotiba GT. A study of orofacial tumors in Nigerian children. J Oral Maxillofac Surg 1996;54:34-8.
8. Sehdev MK, Huvos AG, Strong EW, et al. Proceedings: ameloblastoma of maxilla and mandible. Cancer 1974:33:324-33.
9. Grucia B, Stauffer E, Buser D, Bornstein M. Ameloblastoma of the follicular, plexiform and acanthomatous type in the maxillary sinus: a case report. Quintessence Int 2003;34:311-4.
10. Kramer IR, Pindborg JJ, Shear M. The WHO histological typing of odontogenic tumors. A commentary on the second edition. Cancer 1992;70:2988-94.
11. Anastassov GE, Rodriguez ED, Adam AK, Friedman JM. Case report. Aggressive ameloblastoma treated with radiotherapy, surgical ablation and reconstruction. J Am Dent Assoc 1998;129:847-8.
12. Grunwald V, Le Blanc S, Karstens JH, et al. Metastatic malignant ameloblastoma responding to chemotherapy with paclitaxel and carboplatin. Ann Oncol 2001;12:1409-91.