

Plexiform Ameloblastoma and Reconstruction with Free Fibula Flap: A Case Report

Dr. Anshuman Kumar¹, Dr. Ehsan Z Siddiqui², Dr. Surya Udai Singh³, Dr. Nilotpal Mishra⁴

Department of Surgical Oncology, Dharamshila Narayana Superspecialty Hospital, New Delhi, India¹

Department of Plastic Surgery, Dharamshila Narayana Superspecialty Hospital, New Delhi, India²

Department of Surgical Oncology, Dharamshila Narayana Superspecialty Hospital, New Delhi, India³

Department of Plastic Surgery, Dharamshila Narayana Superspecialty Hospital, New Delhi, India⁴

Abstract

Ameloblastoma is the most common odontogenic tumor. Ameloblastomas are infamous for their invasive pattern of growth and a tendency to recur. Therefore, a definitive diagnosis is of utmost importance in order to adequately manage the disease. This case report illustrates a case of plexiform ameloblastoma and highlights the importance of adequate radical resection followed by esthetic and functionally stable skeletal and soft tissue reconstruction using a microvascular free fibula flap. The article also emphasizes on the radiology and histopathology of the disease along with the various modalities to treat it and the reconstructive options available.

Keywords: Ameloblastoma, Reconstruction, Free fibula flap, Plexiform, Case report

Date of Submission: 20-01-2021

Date of Acceptance: 04-02-2021

I. Introduction

Ameloblastoma is the most common odontogenic tumor and a true neoplasm of enamel organ type tissue. It accounts for approximately 11% of all odontogenic tumors.¹ As defined by Robinson in 1937, Ameloblastoma is an anatomically benign, unicentric, non functional, intermittent in growth and a clinically persistent tumor.² Majority of the cases are diagnosed in third to fifth decades of life, however the lesion may be found in any age group.³ Males and females are affected with equal frequency, however certain studies have shown a male preponderance.^{4,5} Ameloblastomas are infamous for their invasive pattern of growth and the tendency to recur. A definitive diagnosis is of utmost importance in order to adequately manage the disease. This case report presents a study of plexiform ameloblastoma which was managed surgically followed by reconstruction with a microvascular free flap and also emphasizes on the radiology and histopathology of the disease along with the various modalities to treat it and the reconstruction options.

II. Case Report

A 31 year old male presented with a swelling in left cheek since one and a half years.

Extraoral clinical examination of the patient was notable for facial asymmetry and a firm swelling in the area of the left mandibular ramus region extending to the base of the mandible (Figs). Intraorally, the area was slightly tender and the tooth had grade-1 mobility.

A cone beam computed tomography was performed which revealed multilocular, radiolucent lesion in the left mandibular posterior region extending from canine till second molar. It was expansile, well defined with curved borders giving it an appearance of a soap bubble.

Expansion of the labial and lingual cortical plates was noted with perforation and discontinuity at the labial cortex and thinning of the lingual cortex. Extensive root resorption was seen with respect to canine, premolars and first molars. Loss of bone till the base of the mandible was noted. Based on the clinical and radiological findings, a differential diagnosis of ameloblastoma, odontogenic keratocyst, aneurysmal bone cyst of the left mandible was formulated.

An incisional biopsy under conscious sedation was carried out in the operation theatre and the specimen was sent for histopathological examination. The findings revealed the tumor being composed of odontogenic epithelium with cells arranged in the form of anastomosing cords and sheets, along with peripheral basal cells and central stellate reticulum. Thus, a confirmatory diagnosis of ameloblastoma -plexiform type was made. The patient was planned for segmental mandibulectomy followed by microvascular osteomyocutaneous free fibula flap reconstruction.

Surgical resection of the tumor was carried out through an extraoral submandibular approach. Subplatysmal flaps were raised and segmental mandibulectomy was performed. Exploration of the left neck was done to expose the donor vessels for anastomosis. The case was handed over to the plastic surgeon for reconstruction. A free fibula flap was harvested from the patient's left leg with a skin paddle of about 22 x 5 cm based on two perforators. Skeletal reconstruction was done by making osteotomies in the fibula, 6 cm for the body of mandible and 2 cm for reconstructing the symphysis, followed by fixation with 2mm plates and screws. Anastomosis in the left neck was carried out with facial artery, internal jugular vein and external jugular vein as the donor vessels followed by interpolation and inseting of the flap to fill up the defect. Flap vitality was confirmed and the wounds were closed over drains. Broad spectrum antibiotics and analgesics were prescribed for 10 days. The entire post operative period was uneventful with good wound healing. The patient had an excellent recovery both functionally and esthetically. He was under a regular follow-up and after three months, the patient was apparently better with no donor or recipient site complications.

III. Discussion:

Ameloblastoma is one of the most common benign odontogenic tumor.⁶ It comprises 1% of all radiolucent jaw lesions.⁷ Eighty percent of ameloblastomas arise in the mandible, mainly in the molar ramus region.⁸ Histologically, there are two basic patterns, follicular and plexiform, among others such as cystic, desmoplastic, acanthomatous, and granular cell with follicular being much more common.⁹ Ameloblastoma of the mandible has a recurrence rate of 8% in solid/multicystic variant and 4% in unicystic variant.⁶ Plain radiography, panoramic radiographs, computed tomography (CT), magnetic resonance imaging (MRI), and histopathology are all used as diagnostic aids. Radiological findings may include expansion of cortical plates with scalloped margins, multiloculations or 'soap bubble' appearance and/or root resorption.¹⁰ Multicystic ameloblastoma has a higher tendency to recur as compared to unicystic ameloblastoma. The reason for this higher rate is believed to be due to the multiple microextensions the tumor has projecting into the bone.¹¹ Surgery is the mainstay of treatment for ameloblastoma. Treatment ranges from conservative surgery to more radical procedures where the former includes radiotherapy, curettage and enucleation. Radical surgery, as defined by Muller and Slootweg, is a procedure wherein the ameloblastoma is excised with a margin of normal bone.⁸ Most surgeons suggest resecting at least 1 cm of uninvolved bone beyond the tumor margin. When the tumor has extended beyond the bone, inclusion of adjacent soft tissue into the resection specimen must be performed to ensure complete tumor-free soft tissue margins.¹² Sowjanya kalwagadda et al.¹³ conducted a retrospective study to review the management and reconstruction of ameloblastoma of mandible in different age groups over a period of 11 years. They evaluated the outcomes in terms of esthetics, function and choice of reconstruction in different age groups. The various reconstruction options they evaluated included using a reconstruction plate, iliac crest free grafts, and free flaps such as free fibula. They inferred free fibula flaps as the gold standard for mandibular reconstruction.

The free fibula flap has many advantages over other available donor sites for reconstruction of the mandible. It is easier to harvest, more reliable when anastomosis is concerned, and has less postoperative morbidity. Furthermore, it has a thicker cortex than the scapula, radius, or ilium. The fibula is relatively easy to contour and can acquire a solid union with the mandible by a month after transfer.¹⁴

IV. Conclusion :

This case report illustrates a case of plexiform ameloblastoma and highlights the importance of adequate radical resection followed by esthetic and functionally stable skeletal and soft tissue reconstruction using a microvascular free fibula flap. It is imperative to adequately diagnose a case of ameloblastoma and meticulously formulate the treatment plan. Surgical resection with adequate margins remains the modality of choice.

REFERENCES

- [1]. Neville BW, DD D. Allen CM, Bouquot JE. Oral & Maxillofacial Pathology. 3rd ed. St Louis: Saunders Elsevier. 2009:507-23.
- [2]. Robinson HB. Ameloblastoma: a survey of 379 cases from literature. Arch Path. 1937;23:831-43.
- [3]. Kreppel M, Zöller J. Ameloblastoma—Clinical, radiological, and therapeutic findings. Oral diseases. 2018 Mar;24(1-2):63-6.
- [4]. Bansal S, Desai RS, Shirsat P, Prasad P, Karjodkar F, Andrade N. The occurrence and pattern of ameloblastoma in children and adolescents: an Indian institutional study of 41 years and review of the literature. International Journal of Oral and Maxillofacial Surgery. 2015 Jun 1;44(6):725-31.
- [5]. Butt FM, Guthua SW, Awange DA, Dimba EA, Macigo FG. The pattern and occurrence of ameloblastoma in adolescents treated at a university teaching hospital, in Kenya: a 13-year study. Journal of Cranio-Maxillofacial Surgery. 2012 Feb 1;40(2):e39-45.
- [6]. Antonoglou GN, Sándor GK. Recurrence rates of intraosseous ameloblastomas of the jaws: a systematic review of conservative versus aggressive treatment approaches and meta-analysis of non-randomized studies. Journal of Cranio-Maxillofacial Surgery. 2015 Jan 1;43(1):149-57.
- [7]. Regezi JA, Kerr DA, Courtney RM. Odontogenic tumors: analysis of 706 cases. Journal of oral surgery (American Dental Association: 1965). 1978 Oct;36(10):771.
- [8]. Gardner DG. Plexiform unicystic ameloblastoma: a diagnostic problem in dentigerous cysts. Cancer. 1981 Mar 15;47(6):1358-63.

- [9]. Sattar MA, Uzzaman S. Ameloblastoma An asymptomatic odontogenic tumor. *Bangladesh Journal of Otorhinolaryngology*. 2012 Nov 24;18(2):238-41.
- [10]. Minami M, Kaneda T, Yamamoto H, Ozawa K, Itai Y, Ozawa M, Yoshikawa K, Sasaki Y. Ameloblastoma in the maxillomandibular region: MR imaging. *Radiology*. 1992 Aug;184(2):389-93.
- [11]. Müller H, Slootweg PJ. The growth characteristics of multilocular ameloblastomas. A histological investigation with some inferences with regard to operative procedures. *Journal of maxillofacial surgery*. 1985 Oct;13(5):224-30.
- [12]. Williams TP. Management of ameloblastoma: a changing perspective. *Journal of oral and maxillofacial surgery*. 1993 Oct 1;51(10):1064-70.
- [13]. Kalwagadda S, Kumar B, Nair SC, Shah AK, Shroff SS. Management of Ameloblastoma With Free Tissue Flap in Comparison With Other Reconstructive Options Available. *Journal of maxillofacial and oral surgery*. 2020 Jun;19(2):283-8.
- [14]. Guerra MF, Gías LN, Campo FJ, González FJ. Vascularized free fibular flap for mandibular reconstruction: a report of 26 cases. *Journal of oral and maxillofacial surgery*. 2001 Feb 1;59(2):140-4.

FIGURES



Figure 1: Preoperatively swelling is noted on the left side of the face over the mandible



Figure 2: CBCT showing radiolucent lesion in the left mandibular region with perforation and discontinuity at the labial cortex. The lesion has a soap bubble appearance.



Figure 3a



Figure 3b

Figure 3a shows the defect post segmental mandibulectomy. Figure 3b shows the osteotomized free fibula flap that is harvested as an osteomyocutaneous flap.



Figure 4: 3 months post-operatively

Dr. Anshuman Kumar, et. al. "Plexiform Ameloblastoma and Reconstruction with Free Fibula Flap: A Case Report." *IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)*, 20(01), 2021, pp. 41-44.