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IP International Journal of Maxillofacial Imaging

Journal homepage: <https://www.ijmi.in/>

Case Report

Reconstruction of large ameloblastoma crossing midline with free fibula flap- A case report

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ARTICLE INFO

Article history:

Received 11-03-2022

Accepted 15-03-2022

Available online 07-04-2022

Keywords:

Ameloblastoma

Free fibula flap

Mandibular reconstruction

ABSTRACT

Ameloblastoma is the second most common odontogenic tumor of the oral cavity with the primary site being the mandible. It affects more the mandible than the maxilla. The most common complaint is a painless swelling over the mandibular area. Despite its benign nature, ameloblastoma has a high local recurrence rate, with the most recurrences seen within 5 years after operation. Biopsy and radiological evaluation is mandatory. Although this tumor is benign, because of its behavior it is locally aggressive and requires the most often surgical resection. In our case, a male 36-year old reported with ameloblastoma crossing the mid-line. Ameloblastoma was confirmed on radiographic and biopsy report. The lesions crossing the mid-line made the reconstruction more challenging. As taking into consideration the advantages of free fibula flap, our reconstruction was planned using the same flap.

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1. Introduction

In 1827, the first person to identify ameloblastoma was Cusack. A rare benign odontogenic tumor is Ameloblastoma,^{1,2} with its name derived from the early English word “amel” meaning enamel and the Greek word “blastos” meaning germ.³ Odontogenic tumor of the jaw ameloblastoma, is derived from the dental embryonic remnants possibly from the epithelial lining of an odontogenic cyst, dental lamina or enamel organ, stratified squamous epithelium of the oral cavity or displaced epithelial remnants.⁴ About 70% incidence of occurrence is reported in the mandibular molar-ramus region than maxilla by the ratio of 5:1, and it accounts for 1% of all cysts/tumors of jaws and 18% of all odontogenic neoplasms.⁵ Third to fifth decade of life is the average age reported and there is no sex predilection when the tumor is not associated with an unerupted tooth; the gender

ratio is male to female ratio of 1:1.8.⁶ Most of the ameloblastoma which have perforated the cortex i.e buccal or lingual needs to undergo surgical excision. Therefore, in most of the cases surgical management is preferred in Ameloblastoma. Curettage technique has a high recurrence rate and use is therefore discouraged. Wide resection with approximately 1 cm margins with immediate reconstruction of the mandible is usually preferred for its treatment. Commonly used reconstruction technique by the surgeons is vascularized osteocutaneous free flap with plate-screw mandibular reconstruction. For reconstruction of almost the entire segment of the mandibular defect, fibula is the primary source of bone and it has a very good quality of blood vessels.⁷

The first lower-jaw reconstruction with a fibular flap was done in 1989, using osteotomies to mimic the shape of the mandible, was described. The main advantage of micro vascular free flaps is the applicability of these flaps due to their surface texture characteristics, tissue volume, vascular nutrition, and possibility of transmitting the diverse tissues.

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Free fibula flap as bone graft has become a cornerstone in the head-neck armamentarium.⁸ We report a case of ameloblastoma crossing the midline of jaw.

1.1. Case Report

A case of 36-year-old male with a complaint of swelling on the left side of the mandible and crossing the midline and extending toward right side of mandible reported to our department. The swelling is present since 10 – 12 years. Patient had gone to a dentist and had undergone surgical procedure (data not available). Patient also gives history of paresthesia on the side of swelling. He also experienced pus and blood discharge from the same side. Our examination of the patient revealed, extra orally there was hard and tender swelling that extended from the left angle of mandible crossing the midline. Intra orally swelling seen to be extending from tooth number 35 region. Expansion of buccal and lingual cortical plates seen.

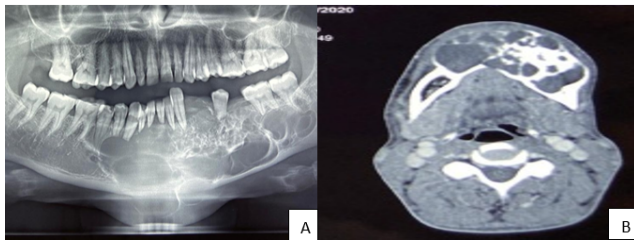


Fig. 1: A: Pre-operative OPG B: CT evaluation

Radiographic evaluation revealed well-defined multilocular radiolucency extending from the left angle of mandible crossing the midline. Histopathological report revealed H and E stained section shows single tiny bit of tissue showing proliferation of odontogenic epithelial islands in fibrocellular stroma. In few islands tall columnar ameloblasts like cells with centrally placed stellate reticulum like cells or squamous metaplasia is noted. Ameloblastic follicle appears more compressed and showing elongated, needle like ends. Overall histopathological features are suggestive of *Acanthomatous Ameloblastoma*. Tracheostomy was planned for the patient. Mandibular resection was performed, segmental mandibulectomy with a surgical margin of 2 cm from the radiographic border of the tumor was performed. The tumor measured approximately 8-9 cm.

Mandibular reconstruction was done immediately with a free fibula osteocutaneous following resection. Leaving 5–7 cm of bone distally and proximally and preserving at least one perforator to the skin, the maximum possible length of fibula bone was harvested. Into two or three segments the bone was osteotomized, which would be attached to a premolded mandibular reconstruction plate with screws after dividing the donor artery and vein. To provide external definition of the jaw the plate was placed

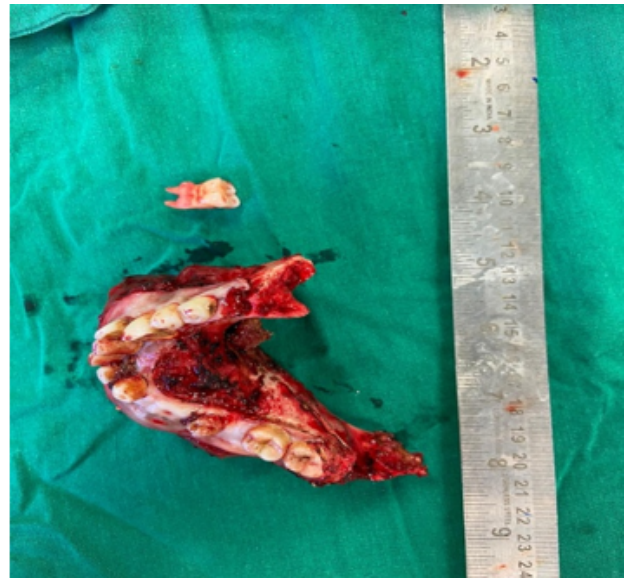


Fig. 2: Tumor was removed in toto



Fig. 3: Fibula flap marking

at the lower border of the mandible. The patient recovery was uneventful postoperatively. The specimen was sent for histopathologic examination then after. The postoperative histopathologic report was consistent with the preoperative report confirming the diagnosis of ameloblastoma. All margins were clear.

2. Discussion

Ameloblastoma is a benign odontogenic tumor which represents 10% of all jaw tumors.⁹ It has the potency to achieve a massive size which can cause facial asymmetry. According to the literature, mandibular posterior region is most frequently encountered site for ameloblastoma and usually diagnosed between 30 and 60 years.¹⁰ In the current case, taking into account the site and history, a provisional diagnosis of ameloblastoma was



Fig. 4: Post-operative OPG

made. Despite of clinical and radiographic suspicion in favor of ameloblastoma, a biopsy is always mandatory in such extensive lesions to exclude malignancy.¹¹

A study was conducted by *Rizzitelli A et al.*¹², it was a population-based study of malignant ameloblastoma. It was carried to determine its incidence rate and absolute survival. Total 293 patients were looked at across the United States. In this study they founded that the overall incidence rate of malignant ameloblastoma was about 1.79 per 10 million persons/year. They also found that malignant ameloblastoma, comprising the two types, metastasizing ameloblastoma, and ameloblastic carcinoma, represents 1.6 to 2.2% of all odontogenic tumors. Previous epidemiologic research were also confirmed through this study, which showed the male to female ratio to be between 2.3 and 5.¹² In *Northern California* a study was carried out according to a retrospective study done for both primary management and treatment of recurrences for mandibular ameloblastoma, specific diagnostic and treatment techniques had been followed which gave satisfactory results.⁵ *Taylor et al.* in 1975 first reported the free vascularized fibula graft. For large posttraumatic defect of the contralateral tibia they successfully used the graft. The vascularized fibula transfer gained in popularity in the ensuing years, and its indications were extended. The fibula was investigated as a donor site for free-flap mandible reconstruction.^{12,13}

Fibular bone allows to plan osteotomies in relation to the orientation of the bone and its vascular pedicle. Cortical bone is thick and it readily accepts plates and screws for a secure interosseous fixation and osteointegrated implants may be placed in this safely. The first choice for the head and neck surgeon is fibular flap which has many advantages for mandible reconstruction than other alternatives like scapular and crista ilaca flaps.¹³ The presence of long bone in the flap is the main advantage of fibula flap that can be used to repair the bone defects. Low morbidity, hidden scar, facilitation of access to further tissue, long vascular pedicle, adequate blood supply, ability to simultaneously transfer the skin, bone, and soft tissue along with one-

stage vascular anastomosis are the benefits of this flap.^{14,15} Taking into consideration these benefits of free fibula flap, in our case as the lesion was large crossing the midline we planned for reconstruction with free fibula flap. In our case reconstruction was necessary to avoid tongue fall, to preserve the continuity of the jaw, and esthetic smile of the patient.

Study conducted by *Haluk Vayvada et al.*, patients with mandibular ameloblastoma, segmental mandibulectomy and immediate reconstruction with fibular flap was performed. The patients were followed up for a mean of 29.3 months (range, 17-38 months). By using a questionnaire the functional and esthetic results were also evaluated. The questionnaire consisted of questions addressing oral continence, diet, social activities, speech, and facial appearance. Is the result was segmental mandibulectomy with safe borders and immediate reconstruction with free fibula flap or deep circumflex Iliac artery flap is an ideal treatment method for mandibular ameloblastoma.¹⁶ *Marlinda Adham*, presented a case of ameloblastoma which was treated with hemimandibulectomy. The defect was reconstructed with a free vascularized fibular graft.⁷

A study conducted by *Hemant and Saraiya*, a record of follow-up ranged from 6 months to 7.7 years with a mean of 5.1 years was maintained. Patients treated with curettage, tumor recurred within a year in all two patients (100%). Out of 35 radical excisions, only one patient (2.85%) developed recurrence 3 years after the disease-free interval. Good mouth opening, intelligible speech, satisfactory lower jaw shape, and facial profiles were achieved in all 29 patients who were treated with primary free fibular flap.¹⁷

3. Conclusion

Poor symptoms and low prevalence results in late diagnosis of ameloblastoma. Resection with safety margins and immediate reconstruction whenever possible is preferred treatment. For better treatment definition, routine histological classification of the ameloblastoma is mandatory for its morphological characterization. The main success factor associated with the treatment is the early diagnosis and to correlate the histopathologic findings with clinical and radiographic features to achieve a correct definitive diagnosis as all such lesions might have prognostically different biologic behaviors and the final diagnosis may alter the therapeutic decision significantly.¹⁸

4. Source of Funding

None.

5. Conflict of Interest

None.

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Cite this article: Lade AY, HS, Sachdeva S, Shah S. Reconstruction of large ameloblastoma crossing midline with free fibula flap- A case report. *IP Int J Maxillofac Imaging* 2022;8(1):16-19.