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Massive Ameloblastoma and Reconstruction with Free Fibular Graft

Linda Jana Sintaningtyas[1*], Joko Purnomo[2] and Affandi Wiramur[3]

[1]Department of Surgery, Faculty of Medicine, Sebelas Maret University, Surakarta, Indonesia.
[2]Division of Surgical Oncology, Department of Surgery, Sebelas Maret University, Surakarta, Indonesia.
[3]Division of Plastic and Reconstructive Surgery, Department of Surgery, Sebelas Maret University, Surakarta, Indonesia.

Authors' contributions

This work was carried out in collaboration among all authors. Author LJS wrote the draft of the manuscript, managed the analyses of the study and literature searches. Authors JP and AW designed the study and did collaboration in operation theatre. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aims: To report a case of massive ameloblastoma in mandible with wide excision and reconstruction with free vascularized fibular graft and titanium plate.
Case Description: A 49- year-old male patient complained right facial mass since 5 years ago. Patient underwent right hemimandibulectomy with general anesthesia and pathologic examination revealed folliculare ameloblastoma. The large defect after resection reconstructed with free fibular graft. Fibula was osteotomised and resembled with mandible shape and reconstructed by plate with intact pedicle. Care was taken to protect the periosteal branch of the peroneal artery before performing an osteotomy. Shaping of the resected fibula was done according to the preoperative template. A titanium miniplate with locking screws was used to secure the osteotomized fibula and the mandible. To secure the airway, we performed tracheostomy. Followup after operation, patient still got a defect facial asymmetry in right mandible.
Discussion: Ameloblastoma is histologically benign but locally aggressive tumor originating from odontogenic epithelium. After hemimandibulectomy, reconstruction continued with microvascular

**Corresponding author: E-mail: dr.lindajana@gmail.com;*

free fibular graft and titanium plate. Osteotomies were performed with the pedicle still attached.
Conclusions: Free vascularized fibular graft with titanium plate is preferrably reconstruction option for large defect after wide excision of mandibular ameloblastoma.

Keywords: Ameloblastoma; reconstruction; free fibular graft.

1. INTRODUCTION

Ameloblastoma may arise from remnants of tooth-forming components, such as rests of dental lamina, developing enamel organ and the epithelial lining of odontogenic (dentigerous) cysts, or possibly from the basal epithelial cells of the oral mucosa [1]. It is an aggressive benign tumour of epithelial origin that has generally been treated surgically. Accurate preoperative imaging can help complete surgical resection with clear margins for long term cure. The most common site for ameloblastoma is mandibular molar region. The management of ameloblastoma is surgical with approximately 1 cm margin.

In this case, we report a patient who underwent right ameloblastoma hemimandibulectomy and reconstruction with free vascularized fibular graft. Before operation, we performed tracheostomy to secure the airway.

2. PRESENTATION OF CASE

A 49-year-old male patient complained right facial mass, which was diagnosed as ameloblastoma from previous biopsy. The lump began to appear in the last 5 years, getting bigger slowly, but the patient never went to the doctor because of fear. The size of mass is like a tennis ball with an ulcer wound on the top. The patient had difficulty chewing solid food and spoke. When the patient came, we did a thorough examination in physical, laboratory and radiological examinations. No abnormalities were found on the laboratory examination. From the ct scan results, it was found that a mass compound with solid, fat, calcified, necrotic and air in it, expansion in the corpus to the right mandibular angle with a soap bubble appearance that infiltrated the masseter muscle, levator anguli oris muscle, pterigoideus medial muscle, hypoglossus muscle, superior constrictor muscle, right pharynx, genioglossus muscle, which pushed against the uvula and oropharynx to the left side as high as VC3-4 level, destroys the corpus to the right mandibular angle and attached to the lateral wall of the right maxillary sinus, and there were multiple lymphadenopathy sublingual bilateral and left submandibular colli.

During the operation, general anesthesia was performed with a nasal endotracheal tube. Then a tracheostomy was performed to maintain patency of the airway. A wide excision was carried out on the tumor mass starting from the right mandibular condylus to the left premolars and the specimen was examined in the Pathology Anatomy laboratorium. Wide excision leaves a defect from right condyle to the left body of mandible, where the palate and inner lanning were still intact. A defect was 8 cm long. The facial artery and jugular vein were identified, then skeletonized and preserved the pedicle along 6cm. Next, the donor was taken from the left fibula bone. The design for molding the fibula was created in accordance with the drawings. Fibular graft was performed (the bone was taken 8cm from the proximal and 8 cm from the distal with preservation of the fibular artery and vein (8cm donor pedicle). Harvested left fibular bone is fixed by reconstructed the mandibular plate from the condyle area. 3 screws were installed and the plate was fixed to the glenoid fossa with a non-absorbable 2.0 thread. The inner lining was sutured with absorbable 4.0 thread to the skin and performed the anastomosis was perfomed end to end artery between facial artery with peroneal/ fibular artery and to end vein of the jugular vein branches with a non-absorbable 9.0 thread with an ischemic time of 45 minutes. Veins are diagnosed first, then arteries. Redon drain than put into the mandible and the outer skin was then sutured with non absorbable 6.0 thread. For donor, fibula was sewn layer by layer and put a drain.

Pathology anatomical of tumor tissue received mandibular tissue sized 16x13x12 cm, that was encapsulated, with a surgical margin 1 cm in the front of the bone, brittle white colour, transverse cutting, with a microscopic image of tumor tissue arranged in islands, that consist of tumor cells at the edges which was arranged palisading and consist of monomorphous columnar cells, and monomorphous stellate cells in the middle. There are also cystic areas. No signs of malignancy are found. The conclusion of the mandibular tissue is folliculare type of ameloblastoma.

Fig. 1. Patient with massive ameloblastoma

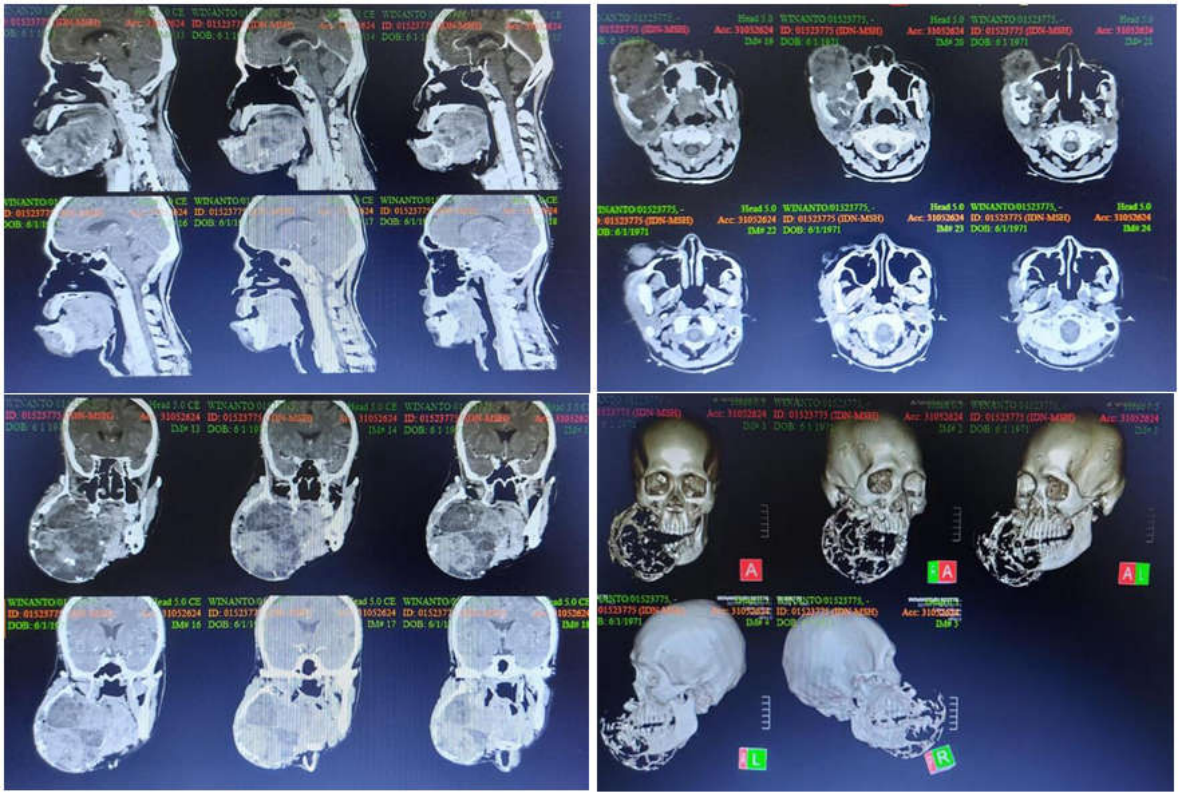

Fig. 2. CT scan pre operation

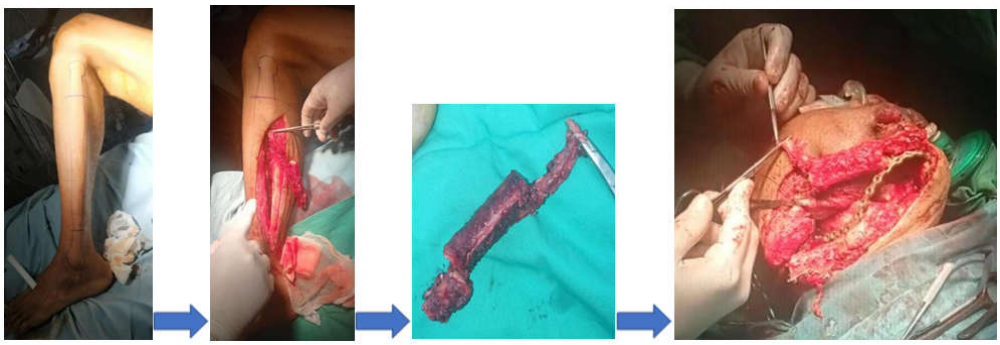

Fig. 3. Durante Operation: Reconstruction with microvascular free fibula graft and titanium reconstruction plate was done next

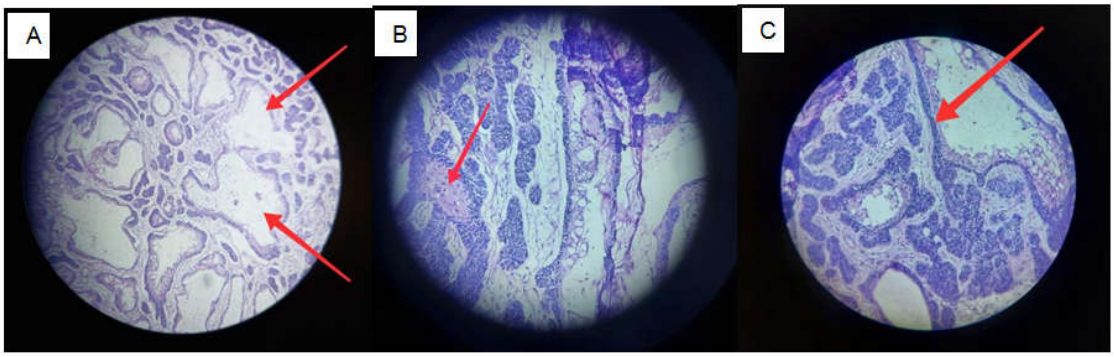

Fig. 4. Histopatologyc examination: A. Cystic area of the tumor B. Squamous metaplasia C. Tumor tissue is arranged in islets consist of tumor cells at the edges which are arranged palisading, consist of monomorphous columnar cells, monomorphous stellate cells in the middle, and cystic area

Fig. 5. Post operation: Above: Day 15th, Below: Day 45th

After surgery, there was still a defect in the right mandible, the face still look asymmetry and right side tongue protrusion anteriorly because right mandibular gingiva was absent.

3. DISCUSSION

Mandibular ameloblastoma can grow massively and cause facial asymmetry. In this case, a 49 years old male patient presented with a mass in the lower right cheek and jaw since 5 years ago which grew slowly and cause facial asymmetry. CT scan revealed a mass compound with solid, fat, calcified, necrotic and air that expand in the corpus to the right mandibular angle with a soap bubble appearance that infiltrated to the muscle.

Patient underwent operation with general anesthesia. Tracheostomy was done first to keep airway patent then continued with right hemimandibulectomy and large defect after wide excision was reconstructed with microvascular free fibula graft and titanium reconstruction plate. The fibula was exposed and a 10 cm segment of fibular bone was cut with oscillating saw. The distal cut was 8 cm from the ankle joint so it was not disturb joint stability. The vascular pedicle was carefully dissected. Osteotomies were performed with the pedicle still attached. Care

was taken to protect the periosteal branch of the peroneal/ fibular artery before performing an osteotomy. Shaping of the resected fibula was done according to the preoperative template. A titanium miniplate with locking screws was used to secure the osteotomized fibula and the mandible. Postoperative histopathologic examination revealed a follicular type of ameloblastoma.

Ameloblastoma is classified as a benign odontogenic tumour, although its behaviour is similar to that of true neoplasia for being locally invasive and aggressive. Historically multicystic or solid ameloblastoma are treated with curettage and enucleation. Normally if the tumour is small (<5cm), the defect can be repaired with a free bone graft [2]. However if the tumour resection is large, reconstruction is challenging and may require a microsurgical flap either from fibula, iliac crest, scapula, radius or ribs to achieve function, cosmetic and restoration of mastication and speech to enhance the quality of life. Treatment by wide excision is curative in up to 95% of cases. Chana et al proposed a 1-2 cm normal margin, and the large defect left after resection can be reconstructed preferably with the fibula osteocutaneous flap [3]. Taylor in 1975 first described vascularised fibula graft for composite reconstruction of the bone and soft tissue defect. After osteotomies, it performed vascularised fibula grafts. The free fibula flap provides the greatest bone length and is suitable to accept dental implants. Osseointegrated implants have become generally accepted for prosthodontic management. The application of endosseous implants in combination with bone grafting for jaw reconstruction has allowed for improved results. Different types of osseointegrated implants have been placed simultaneously with bone grafts or at a later stage after the bone grafts have healed [4].

According to World Health Organization (WHO) 2005 classification of odontogenic tumors, ameloblastoma is divided into four categories: (a) solid/multicystic, in which locally invasive tumor will infiltrate through the medullary spaces and may show multicystic lesions; (b) unicystic, presenting as a cystic intraosseous growth pattern, which is observed clinically and radiographically; (c) peripheral, which is identical to the intraosseous ameloblastoma but appears exclusively in the oral mucosa (extraosseous); and (d) desmoplastic, an infiltrative intraosseous tumor characterized by extensive stromal collagenization or desmoplasia, radiographically appearing as a radiolucent–radiopaque lesion mimicking a fibro-osseous lesion [5].

The most common histopathologic patterns in ameloblastoma are follicular and plexiform patterns. Other microscopic patterns include acanthomatous, granular, and basal cell. These patterns can be uniform or mixed [6]. Surgery is the first choice of treatment of ameloblastoma. The treatment of ameloblastoma can be divided into conservative treatment (enucleation, curettage, and cryosurgery) and radical treatment (marginal or segmental resection) [7].

Ameloblastoma is generally not radiosensitive, but very radioresistant [8]. Ameloblastoma has a high recurrence rate when treated other than mandibular resection [9]. Because of invasiveness, clinically malignant tumor, the rational treatment of ameloblastoma is complete surgery.

4. CONCLUSION

The vascularised free fibula graft provides a good alternative than the other bone grafts for mandible reconstruction. It is relatively easy to perform and carries a low complication rate. The most important to treatf ameloblastoma is completeness removal of the tumor with margin 1-2cm to prevent recurrence. The challenge to manage a massive ameloblastoma is not only the complete removal of the tumor, but also consider the best technique to reconstruct the mandible, fill the defect, and restore function and cosmetics of face with fibular graft.

CONSENT AND ETHICAL APPROVAL

As per university standard guidelines, participant consent and ethical approval have been collected and preserved by the authors.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Dhanuthai K, Chantarangsu S, Rojanawatsirivej S, Phattarataratip E, Darling M, Jackson-Boeters L, Subarnbhesaj A. Ameloblastoma: A multicentric study. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 2012;113(6):782–788. Available:https://doi. org/10.1016/j.oooo.2012.01.011
2. Campbell D, Jeffrey RR, Wallis F, Hulks G, Kerr KM. Metastatic pulmonary ameloblastoma: An unusual case. British Journal of Oral and Maxillofacial Surgery. 2003;41(3):194–196. Available:https://doi. org/10.1016/S0266-4356(03)00046-9
3. Chana JS, Chang YM, Wei FC, et al. Segmental mandibulectomy and immediate free fibula osteocutaneous flap reconstruction with endosteal implants: an ideal treatment method for mandibular ameloblastoma. Plast Reconstr Surg. 2004;113:80e87.
4. Faqi Nurdiansyah Hendra, Ellen M. Van Cann, Marco N. Helder, Ruslin M, Jan G de Visscher, Tymmour F, Henrica. Global incidence and profile of ameloblastoma: A systematic review and meta-analysis. Oral Disease; 2019. DOI:10.1111/odi.13031. Available:https://wileyonlinelibrary.com/journal/odi
5. Milman T, Ying GS, Pan W, LiVolsi V. Ameloblastoma: 25 year experience at a single institution. Head and Neck Pathology. 2016;10(4):513–520. Available:https://doi.org/10.1007/s12105-016-0734-5
6. Fregnani ER, da Cruz Perez DE, de Almeida OP, Kowalski LP, Soares FA, de Abreu Alves F. Clinicopathological study and treatment outcomes of 121 cases of ameloblastomas. International Journal of Oral and Maxillofacial Surgery. 2010; 39(2):145–149. Available:https://doi.org/10.1016/j.ijom.2009.11.022
7. Santos Tde S, Piva MR, Andrade ES, Vajgel A, Vasconcelos RJ, Martins-Filho PR. Ameloblastoma in the Northeast region of Brazil: A review of 112 cases. Journal of Oral and Maxillofacial Pathology. 2014;18(Suppl 1):S66–71. Available:https://doi. org/10.4103/0973-029X.141368
8. Steal MM, Dib LL, Pinto DS. Management of solid ameloblastoma in the jaw with liquid nitrogen spray cryosurgery, Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 1997; 84(4):339–344.
9. SR. Penanganan ameloblastoma mandibula. Warta IKABI. 1997;10:237–243.

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