

Evaluation Treated of Marginal Resection Followed by Non-Vascularized Fibular Grafts in Ameloblastoma of Mandible: A Case Series

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Abstract

Ameloblastoma is a rare odontogenic tumour of the mandible and maxilla. Although, reconstruction with free flaps remains gold standard, many factors must be considered in this method including the need of surgical expertise and equipment, increased intra operative time, post-operative stay and socioeconomic reasons. Non-vascularized bone grafts from the fibula offers many advantages and could be an alternative method. This case series will present two cases of ameloblastoma treated with radical resection followed by NVFG reconstruction. Information was obtained through patient follow-up, and medical record. Outcome evaluation will be revealed and discussed. The patients were 44 y.o. male and 25 y.o. female. There was no serious complication found due to operation such as infection and wound dehiscence in both patients. A month after surgery, the lump is still existed but only consist of soft tissue swelling. Despite the full normal function have not return yet, the basic function of jaw such as swallowing, and mouth opening are good. Radiological finding a month after operation shows little callus formation in both patients. The radical surgical option is the current standard of care for ameloblastoma while not many studies have been conducted to compare vascularized and non-vascularized bone reconstruction. NVFG can provide a better quantity of bone to replace implants and a more satisfactory contour than vascularized graft. However, NVFG will undergo remodelling process after reconstruction, therefore implant placement would end in failure if inserted too early when the remodelling process has not finished. Vascularized bone graft is indicated in radiation patients while non-vascularized graft is used in non-radiation patients. A good functional outcome and least donor morbidity could be seen in our patient undergone NVFG reconstruction after radical resection. NVFG is a good alternative in small segmental defect after resection of ameloblastoma of the mandible.

Keywords: non-vascularized bone grafts; ameloblastoma.

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1. Introduction

Ameloblastoma is a rare odontogenic tumour of the mandible and maxilla. It is benign but locally aggressive and could cause severe fascial disfigurement, functional disability, and high recurrence rates if treated improperly [1,2]. Ameloblastoma is the most common benign odontogenic tumour in China and Africa, and the second most in the United States and Canada. The estimated incidence is 0.5 cases per million-person years and most diagnosed in the age of 30-60 years. Both men and women have the same incidence according to several studies [2,3].

Ameloblastoma can be clinically classified into unicystic, solid/ multi cystic, peripheral, and malignant.[4] This tumour primarily located in the mandible (about 80% of cases) and the rest of it in the upper jaw, predominantly in the canine or molar region. The clinical findings are often asymptomatic and presented as an incidental finding on imaging examination. However, when symptoms are present, patients often have a slow-growing, painless swelling [2]. Pain, paraesthesia, malocclusion, ill-fitting dentures, teeth loosening, periodontal ulceration, fistula, and nasal airway obstruction are less common symptoms presented [2,4].

Preoperative diagnostic evaluation includes imaging and biopsy. Ameloblastoma, apart from the peripheral subtype, arises within bone thus is often detected incidentally on dental X-rays (pan tomography) or plain radiograph. Soap bubble appearance is seen in most cases of multilocular/ solid type. Lytic lesion with scalloped margin, resorption of tooth roots, and impacted molars can be seen in unicystic type [2]. Computed tomography (CT) may be useful to show the extent of the tumour and destruction of cortex [1]. Histopathological examination was done after an incisional open biopsy or fine needle aspiration of the tumour. Ameloblastoma resembles normal odontogenic/ enamel epithelium and ectomesenchyme [2].

Management of ameloblastoma presented with many challenges. Successful treatment requires both adequate resection and an acceptable functional and aesthetical reconstruction of the defect [1]. Surgery is the standard of treatment in ameloblastoma. Surgical options known for ameloblastoma are conservative and radical surgery. Conservative surgery includes enucleation/ curettage of the bone cavity which could be done in an outpatient basis [2]. However, recurrence rates as high as 75-90% were reported after this type of procedure [4]. The radical surgical option is the current standard of care for ameloblastoma [2,3]. The resection in ameloblastoma is recommended 1-1.5 cm bone margin for unicystic and 1.5-2 cm for solid/ multi cystic type to increase cure rates. Bone margin is the distance away from radiographic expected disease free and oncological safe osteotomy [2]. Discontinuity from resection often severely compromise mastication, speech, protection of airway, and facial aesthetic that makes mandibular reconstruction essential to be performed [5].

Reconstruction options for mandible ranging from metallic reconstruction plate to vascularized bone flaps and are highly individualized according to size and location of mandibular defect, quality of dentition, and donor site morbidity [5]. Bone often used for grafts including the scapula, vascularized and non-vascularized iliac crest, rib, radius, and fibula [6]. Both vascularized and non-vascularized bone grafts could be used with different range of satisfactory results [7]. Although, reconstruction with free flaps remains gold standard, many factors have to be considered in this method including the need of surgical expertise and equipment, increased intra operative

time, post-operative stay and socioeconomic reasons [5]. Non vascularized bone grafts from the fibula offers many advantages and could be an alternative method.

Non vascularized fibula graft (NVFG) is a favourable option for mandibular defects of 6-10 cm under optimum conditions especially in limited socioeconomic status [5]. NVFG create a better contour and bone volume for facial aesthetics and implant insertion. This method is effective when the patient has not been previously irradiated [6]. The healing process of NVFG is by creeping substitution of bone. The graft is replaced by newly formed bone from postcondition and osteoinduction after resorption process. Healing is by phase I of new osteoid formation (approximately 4 weeks) and phase II of bone formation by the effect of bone morphogenic protein (BMP) transforming pluripotent host cell into osteoblastic cells (6 weeks until 6 months) [5].

A study by Foster and his colleagues comparing the outcome of 26 NVFG reconstructions and 49 vascularized bone flaps, found that the incidence of bony union was higher, fewer operations to achieve union, and the implant success rate was significantly greater in NVFG patients[7]. This study is accordance with a randomized control trial by Agrawal and his colleagues in 20 patients with benign mandibular pathology treated by NVFG and iliac bone crest grafts. The study showed that NVFG is as better as iliac crest for reconstruction of mandibular in function, aesthetics, and donor site complications [8].

This case series will present two cases of ameloblastoma treated with marginal resection followed by NVFG reconstruction. Outcome evaluation will be revealed and discussed.

2. Cases Illustration

2.1. Case 1

We reported a 44-years old man suffering an ameloblastoma of right mandible. Firstly, nine months before admission, the patient complained about recurrent bleeding on his gingiva without lump as well as pain. Three months later, he complained a lump that shown if he really noticed it. There was no pain occurred on the lump or other location on his jaw. Subsequently, he came to the hospital in order to get more information about the symptom. In the hospital, he got an x-ray examination (Figure 1), and the doctor said that he had a tumour on his jaw. The lump became bigger time by time and the doctor suggested a CT Scan procedure for diagnosis confirmation (Figure 2) and an open biopsy procedure to reveal the tumour type. Physical Examination shows a diffuse lump like a swelling on the lower jaw with no sign of infection or inflammation such as hyperaemia, discoloration, or abscess production. There was a facial deformity especially on the jaw that shows an enlargement of right jaw. There was loose of one adjacent tooth on the side of lesion, no ulcer or periodontal disease found. The patient felt no pain or tenderness according to the lump, and it was firm on palpation.

X-ray examination reveals a “soap bubble” appearance on the right mandible with well-demarcated border and no sign of matrix calcification. There was no erosion of the tooth roots as well as no extension through the cortex into adjacent soft tissue (Figure 1).



Figure 1: pre-operative x-ray of case 1

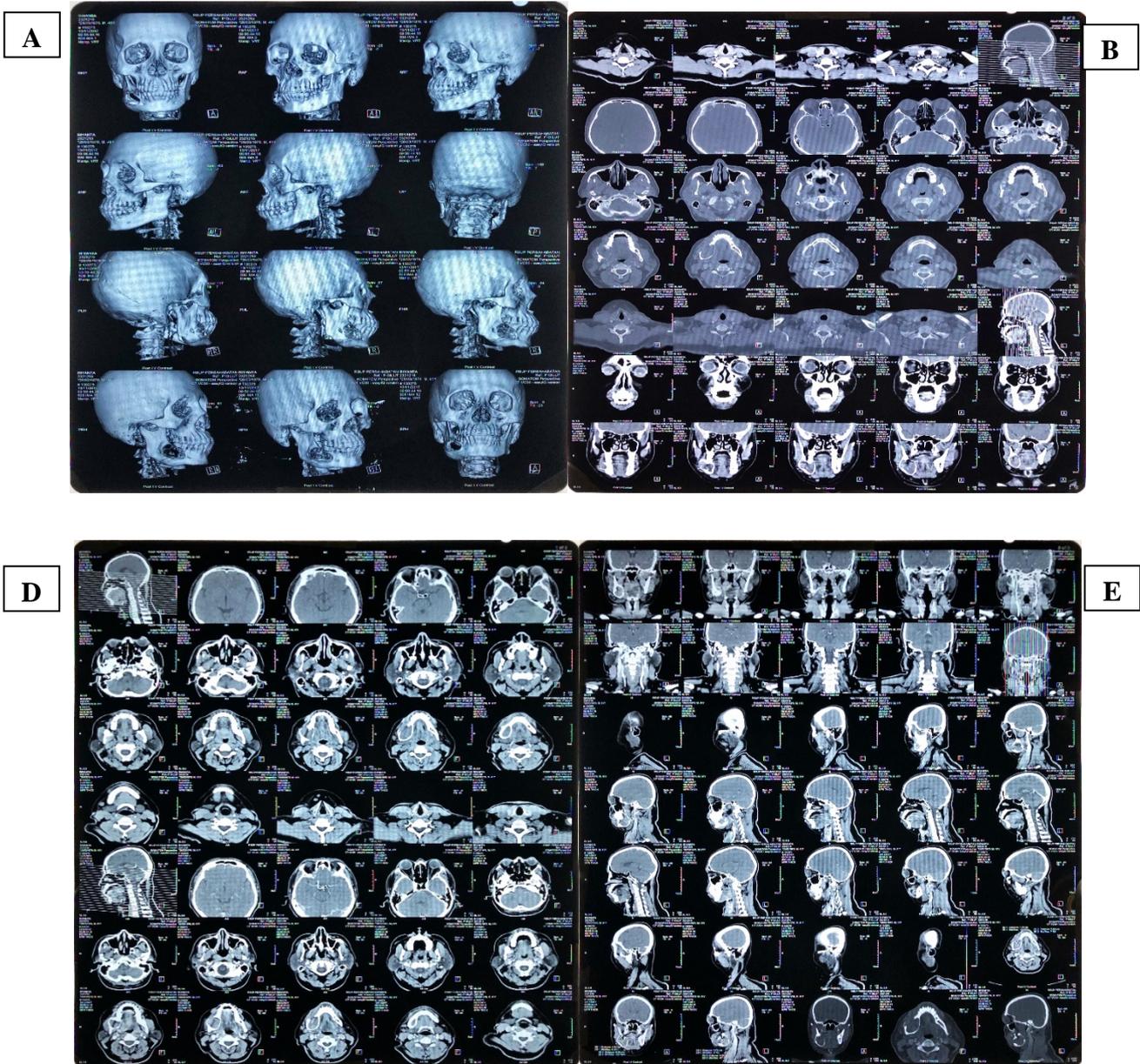


Figure 2: pre-operative CT Scan of case 2

An open biopsy procedure of the tumour showing rounded/ oval odontogenic cell tumour tissue which arranged plexiform, follicular, and palisading with hyperchromatic area on the border of nucleus. There were rough chromatin and eosinophilic cytoplasm. This histological finding supports the diagnosis of plexiform and follicular ameloblastoma.



Figure 3: 1-year follow-up of case 1 showing a good clinical appearance

We choose radical surgery management for this patient. We did a wide marginal resection of the tumour. Before the surgery held, we prepared the plate which will be used for fixation. After identifying the tumour size and location pre-operatively, we incised the overlying skin, and unveil the soft tissue until the bone exposed. We resected the bone with 7 cm length that consist of tumour itself and 1 cm free margin on each side. The non-vascularized fibular graft harvested in similar length with the resected bone (7 cm) from the right leg in order to filling the defect. After bone resection and graft harvesting, the non-vascularized fibular graft is inserted on the bone defect, then fixed with prepared plate and screw. To add more stability, we use a mesh wrapping the graft and then filled it with synthetic bone graft (Figure 4).



Figure 4: post-operative panoramic x-ray, 3month (A), 6 months (B)

2.2. Case 2

We presented a 25-years old female, with chief complain of lump in the left mandible in the past 1 year. Swelling was getting bigger without pain in different location of the jaw. Bleeding occurred both times in 5 and 3 months before admission with tooth loosening. No paraesthesia or other dental symptoms were found. The patient went to our hospital for further information about the symptom.

Physical Examination shows a diffuse lump like on the lower jaw with no sign of infection or inflammation such as hyperaemia, discoloration, or abscess production (Figure 5). There was a facial deformity especially on the jaw that shows an enlargement of left jaw. There was loose of one adjacent tooth on the side of lesion, no ulcer or periodontal disease found. The patient felt no pain or tenderness according to the lump, and it was firm on palpation.



Figure 5: pre-operative clinical appearance of the jaw

X-ray examination reveals a “soap bubble” appearance on the left mandible with well-demarcated border and no sign of matrix calcification. There was no erosion of the tooth roots as well as no extension through the cortex into adjacent soft tissue.

On CT Scan of mandible with contrast, there was soft tissue mass, with non-well-demarcated border, slightly contrasting, no sign of tooth subluxation, no sign of bone destruction, and no sign of intracranial infiltration (Figure 6)

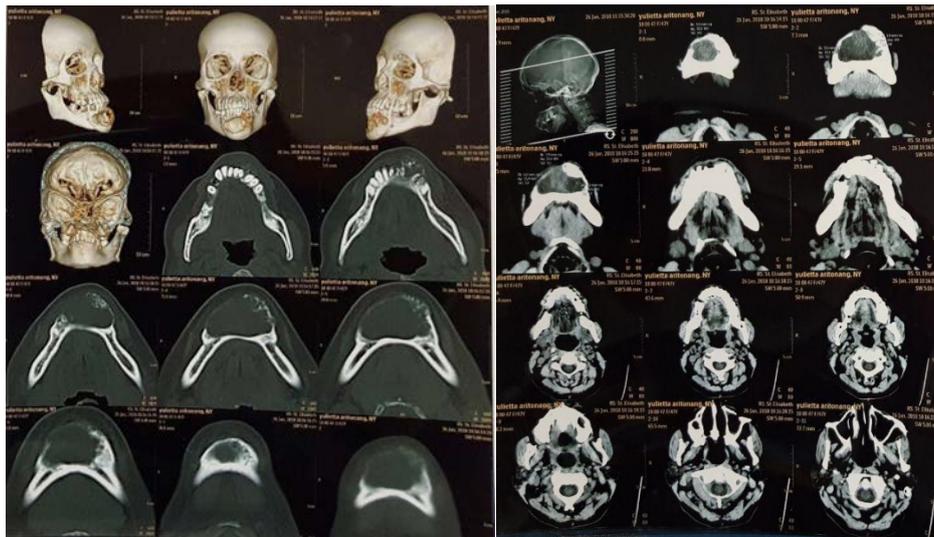


Figure 6: pre-operative MRI

An open biopsy procedure performed on the tumour showing rounded/ oval odontogenic cell tumour tissue which arranged plexiform, follicular, and palisading with hyperchromatic area on the border of nucleus. There were rough chromatin and eosinophilic cytoplasm. This histological finding supports the diagnosis of plexiform and follicular ameloblastoma.

The patient was performed wide marginal resection of the tumour. Before the surgery was held, we prepared the plate which will be used for fixation. After identifying the tumour size and location pre-operatively, we incised the overlying skin and soft tissue. Marginal mandibular nerve was preserved then the facial vein and artery were ligated. The tumour was exposed, and periosteum was stripped. We resected the bone with 8 cm length that consist of tumour itself and 1 cm free margin on each side. The non-vascularized fibular graft harvested in similar length with the resected bone (8 cm) from the right leg in order to fill the defect. After bone resection and graft harvesting, the non-vascularized fibular graft is inserted on the bone defect, then fixed with prepared plate and screw.

3. Results

Table 1: The results of two patients diagnosed with ameloblastoma and treated by wide radical resection and NVFG reconstruction method.

No	Demographical Data	Chief Complain	Diagnosis	Treatment Procedure	Outcome
1.	Male, 44 yo	Recurrent bleeding on gingiva and lump	Ameloblastoma of the right mandible	Wide radical resection and reconstruction using NVFG	No complication Good basic jaw function
2.	Female, 25 yo	Lump of the left mandible	Ameloblastoma of the left mandible	Wide radical resection and reconstruction using NVFG	No complication Good basic jaw function

No serious complication found due to operation such as infection and wound dehiscence in both patients. A month after surgery, the lump is still existed in both cases, but only consist of soft tissue swelling. Despite the full normal function have not return yet, the basic function of jaw such as swallowing is good. Radiological finding a month after operation shows little callus formation in both patients (Figure 4 and Figure 7).

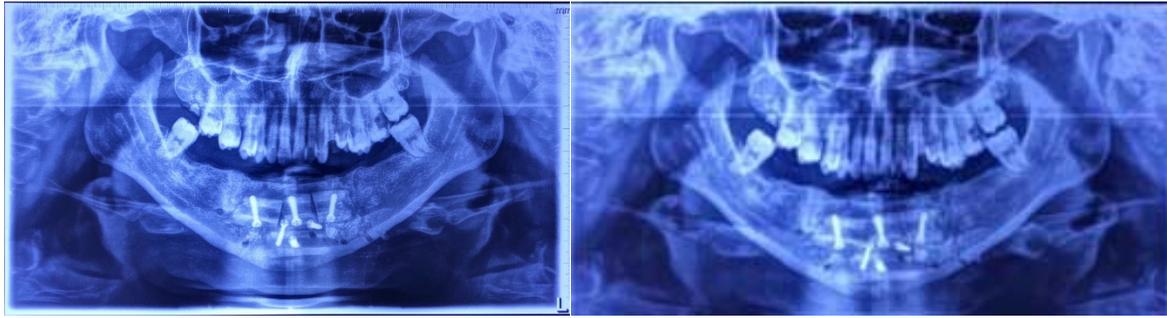


Figure 7: post-operative x-ray of 3 months and 6 months

Table 2: The outcome of jaw function in 3 and 6 months follow-up after wide radical resection and reconstruction using NVFG

No	Demographical Data	Outcome	
		3 months	6 months
1.	Male, 44 yo	Good chewing, soft food eating	solid foods eating, 3 fingers wide mouth opening
2.	Female, 25 yo	Good chewing, soft food eating	solid food eating, good chewing

The jaw function evaluated in 3 and 6-months post-operative was good (Figure 3 and Figure 8). Mouth opening and chewing mechanism was normal. Both of the patients were able to eat solid food 6-months after the surgery.

4. Discussion

NVFG reconstruction remains an accepted alternative method for mandibular defect. Fibula, located in posterolateral of tibia, has a tubular shape similar to mandible. Its dense cortical surface and endosteal blood supply allows multiple osteotomies without compromising viability. It is not a primary weight bearing bone hence its removal will not affect the function of the lower extremity [5].

Not many studies have been conducted to compare vascularized and non-vascularized bone reconstruction. NVFG can provide a better quantity of bone to replace implants and a more satisfactory contour than vascularized graft. However, NVFG will undergo remodelling process after reconstruction, therefore implant placement would end in failure if inserted too early when the remodelling process has not finished. In contrary, vascularized bone undergo very little remodelling but in many cases the quantity of bone is not satisfactory [6].

Many publications stated that NVFG was recommended for graft 6-10 cm. Defects exceeding 9 cm increases the risk of failure using NVFG. Vascularized bone graft is still recommended in larger and longer segmental defect [6,7].

A study by Pogrel and his colleagues comparing 39 patients treated with vascularized bone graft and 29 patients with non-vascularized bone graft showed an additional hospital stay of 14 days and operation time of 3 hours in the vascularized bone graft patients. This result may be caused by difference in age group and history or medical status [6].

Functional and quality of life outcome between vascularized bone graft and NVFG are similar according to many studies. Prior radiation therapy also affects the result with worst functional outcome in the radiation patient. Vascularized bone graft is indicated in radiation patients while non-vascularized graft is used in non-radiation patients. A prospective study by Vu and his colleagues comparing the quality of life evaluation of vascularized versus non-vascularized bone graft reconstruction showed that no significant difference was found in the two groups. However, functional evaluation (chewing and swallowing) showed a favourable results in the non-vascularized group compared to vascularized group [9].

Common complications found in mandibular reconstruction procedure are intra oral wound infection and graft failure. A retrospective study by Devireddy and his colleagues in 7 patients undergone NVFG reconstruction found no complication and a good functional outcome of tongue and jaw movement after 3, 6, and 9 months post operatively [5]. In accordance to other studies, no complication was found in our patient.

A good functional outcome and least donor morbidity could be seen in our patient undergone NVFG reconstruction after radical resection. No prior radiation in ameloblastoma patient also contributed to the favourable result of outcome. NVFG is a good alternative in small segmental defect after resection of ameloblastoma of the mandible.

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