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The sequential treatment of extensive mandibular ameloblastoma

Ruixiu Cheng[1], Xingmao Fu[2], Jianhui Ma[1], Shuliang Li[2], Jie Yu[1], Kun Yan[1], Yang Xue[3*]

[1]Department of Stomatology, The 89th Hospital of PLA, Weifang, China
[2]Institute of Traumatic Orthopedics, The 89th Hospital of PLA, Weifang, China
[3]Department of Oral Biology, School of Stomatology, The Fourth Military Medical University, Xi'an, China;
[*]Corresponding Author: xueyangfmmu@live.cn

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ABSTRACT

Ameloblastoma is the most frequent odontogenic tumor of the jaw. If not treated, ameloblastoma can gain an enormous size and cause severe facial disfigurement and functional impairment. Here we report two patients afflicted with extensive mandibular ameloblastoma (sized in 23 cm × 18 cm × 17 cm and 15 cm × 12 cm × 10 cm, respectively). Both the patients received the same sequential treatment including radical tumor resection, simultaneous reconstruction with fibula free flap graft, vertical distraction osteogenesis on the fibula graft, placement of endosseous dental implants, and final prosthodontic rehabilitation. It took about 15 months to finish the entire course of treatment. And after the four-year follow-up, neither soft tissue related, nor hard tissue related problems were observed. Satisfactory facial symmetry, chewing and speech functions of the patients were restored. So this sequential treatment for extensive mandibular ameloblastoma can obtain an excellent effect by the shortest time and the lowest economical cost. Furthermore, the series also can be used to reconstruct giant mandibular defects caused by different reasons.

Keywords: Ameloblastoma; Mandibular Defect; Fibula Flap; Sequential Treatment

1. INTRODUCTION

Ameloblastoma, a benign but locally aggressive tumor of odontogenic epithelium, is the most frequent odontogenic tumor of the mandible and maxilla [1]. It accounts for about 1% of all tumors and cysts of the jaw and about 10% of the odontogenic tumors. Ameloblastoma is mainly encountered during the third to the fifth decade of life, with equal sex predilection [2,3].

According to different clinical and pathological manifestations, ameloblastomas are typically classified into 3 categories: multicystic/solid, unicystic and peripheral/extraosseous ameloblastomas [2,4]. Multicystic ameloblastoma is the most common variety with an incidence between 63.1% and 85% [4-6]. Unicystic ameloblastoma is a less encountered variant with an incidence between 5% and 15%. It refers to those cystic lesions that show clinical and radiographic characteristics of an odontogenic cyst but histopathologically show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation [7,8]. Peripheral ameloblastoma presents 1.3% to 10% of the cases, showing histological characteristics of intraosseous ameloblastoma that occur solely in the soft tissues covering the tooth-bearing parts of the jaws [2].

The treatment of ameloblastoma mainly relies on surgical operation. However, different operations depend on different tumor types. Peripheral ameloblastoma can be treated with conservative approaches such as enucleation, while unicystic ameloblastoma can be treated with curettage [9]. However, multicystic ameloblastoma requires radical extensive excision. Conservative treatments for this type often lead to recurrence rates between 75% and 90% [4-6]. If not treated, the tumor can gain an enormous size and cause severe facial disfigurement and functional impairment.

The aim of the present report is to show the results of a sequential treatment of patients with huge multicystic ameloblastoma. The sequential treatment includes radical tumor resection, simultaneous reconstruction with fibula free flap graft, distraction osteogenesis (DO), placement of endosseous dental implants and final prosthodontic rehabilitation.

2. CASE PRESENTATION

2.1. Case 1

A 46-year-old male patient presented to our department with an about 23 cm × 18 cm × 17 cm mass in the body of mandible (**Table 1**, **Figures 1(A)-(C)**). Lateral cephalograms and computerized tomographic (CT) scanning with 3 dimension reconstruction demonstrated a great radiolucent region in the mandibular area including the body and ramus (**Figures 2(A)** and **(B)**). After a sequence of treatments, including radical tumor resection, simultaneous reconstruction with fibula free flap graft, DO and dental implantation, satisfied appearance and mandible function were achieved (**Figures 1(D)-(I)**). At the end of the 4-year follow-up, there was no evidence of tumor recurrence and integration of grafted tissue, steady levels of bone around the fixtures, healthy peri-implant tissues and satisfied occlusioon were found (**Figure 3**).

Figure 1. Clinical manifestations of Case 1. (A)-(C): Preoperatively, the patient showed an about 23 cm × 18 cm × 17 cm mass in the body of mandible in anterior view or lateral view. (D)-(F) Two weeks after tumor resection and simultaneous reconstruction with fibula flap, the patient's appearance was well restored; (G)-(I) The patient's appearance after final prosthodontic rehabilitation.

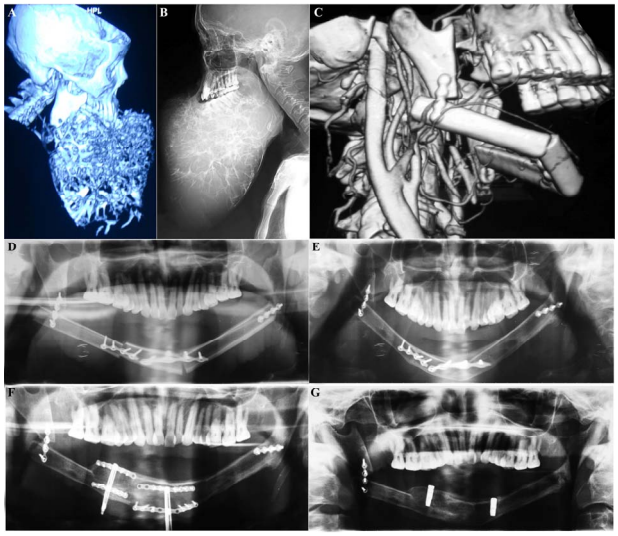

Figure 2. Radiological findings of Case 1. Preoperative CT scanning with 3 dimension reconstruction (A) and lateral cephalogram (B) demonstrated a great radiolucent region in the mandibular area including the body and ramus. CT angiography (C) at 2 weeks after tumor resection and reconstruction with fibula flap showed smooth flow in transplanted peroneal artery and direct plate fixation between the lower ends of the osteotomy in right ramus and fibula bone cortex. The panoramic radiograph (D) at 2 weeks after fibula reconstruction showed obvious sutures at osteotomy sites. Six months after fibula reconstruction, the panoramic radiograph (E) showed good bone healing at osteotomy sites unless the site between the lower ends of right ramus and fibula bone cortex. After three-month consolidation, the panoramic radiograph (F) showed good osteogenesis in the distraction gap. The last panoramic radiograph (G) showed placement of dental implants in the distracted fibula graft.

Table 1. The general information of the two patients.

	Case 1	Case 2
Gender	male	Male
Age (year)	46	47
Tumor growth time (year)	18	3
Rapid growth time (year)	2	1
Tumor volume (cm^3)	23 × 18 × 17	15 × 12 × 10
Tumor weight (kg)	2.7	0.9

2.2. Case 2

A 47-year-old male patient presented to our department with facial asymmetry and an about15 cm × 12 cm × 10 cm mass mainly in the left side of mandible (**Table 1**, **Figures 4(A)-(C)**). CT scanning with 3 dimension reconstruction showed a great multicystic bulging mass mainly in the left mandibular area including the body and ramus (**Figure 5(A)-(D)**). After the same sequence of treatments, satisfied appearance and mandible function were achieved (**Figure 4(D)-(I)**).

Figure 3. Clinical and radiological findings of Case 1 at the end of the 4-year follow-up. (A)-(D) Appearance of the patient at the end of the 4-year follow-up. (E)-(G) Satisfactory occlusion was achieved after placement of implants and prosthetic restoration. H: The intraoral photo showed the soft tissue conditions. (I)-(K) The radiological findings showed integration of grafted tissue, steady levels of bone around the fixtures and osseointegration of dental implants.

3. THE SEQUENTIAL TREATMENT

A treatment panel composed of oral and maxillofacial surgeons, orthopedists, anaesthetists and dentists were established preoperatively. And the same sequential treatment (**Table 2**), including radical tumor resection, simultaneous reconstruction with fibula free flap graft, distractor implantation, DO, distractor removal, placement of endosseous dental implants and prosthodontic rehabilitation, was applied to both the two patients.

3.1. Step 1 Tumor Resection

Under general anesthesia, gross total removal of the tumor was performed by oral and maxillofacial surgeons. In both the two patients, the condylars had not been invaded by the tumor, so the condylars did not be removed in the tumor resection (**Figures 2** and **5**).

3.2. Step 2 Reconstruction with Fibula Free Flap Graft

Preoperatively, mandible plaster model was made according to the proportion of the upper and lower jaw and CT findings. The length of the fibula flap and osteotomy position was determined according to the lower edge of the plaster model. Orthopedists were responsible for cutting the fibula flap, while the oral and maxillofacial surgeons participated in its shaping (**Figure 6(A)**). In order to reconstruction the height of mandibular ramus, direct plate fixation was made between the lower ends of the osteotomy in ramus and fibula bone cortex (**Figure 2(C)**). Then microsurgical vascular anastomosis was made. The oral mucosa and the skin incision were primarily closed. Postoperative course was uncomplicated. CT angiography at 2 weeks after operation showed smooth flow in transplanted peroneal artery (**Figure 2(C)**).

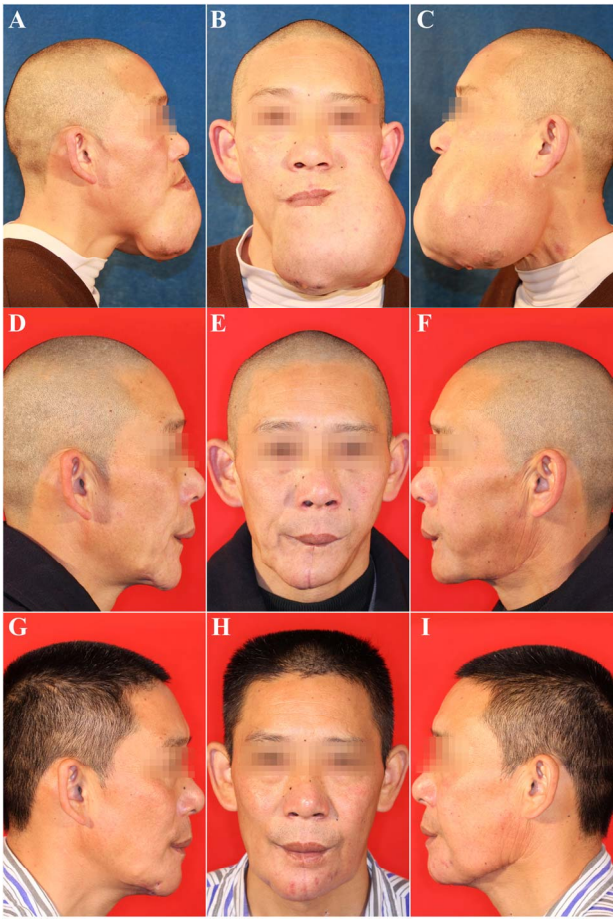

Figure 4. Clinical manifestations of Case 2. (A)-(C) Preoperatively, the patient showed an about 15 cm × 12 cm × 10 cm mass in the left side of mandible in anterior view or lateral view. (D)-(F): Six months after tumor resection and reconstruction with fibula flap, the patient's appearance was well restored. (G)-(I) The patient's appearance after final prosthodontic rehabilitation.

Table 2. The sequential treatment list.

	Treatment	Interval between treatments
Step 1	Tumor resection	
Step 2	Reconstruction with fibula free flap graft	Simultaneous with step 1
Step 3	Distractor implantation	6 Months after step 2
Step 4	Distraction osteogenesis	1 Week after step 3
Step 5	Distractor removal	3 Months after step 4
Step 6	Placement of endosseous dental implants	3 Months after step 5
Step 7	Prosthodontic rehabilitation	3 Months after step 6

3.3. Step 3 Distractor Implantation

Six months later, the patients received distractor implantation under general anesthesia. When the mandible was exposed, optimal integration of grafted tissue and

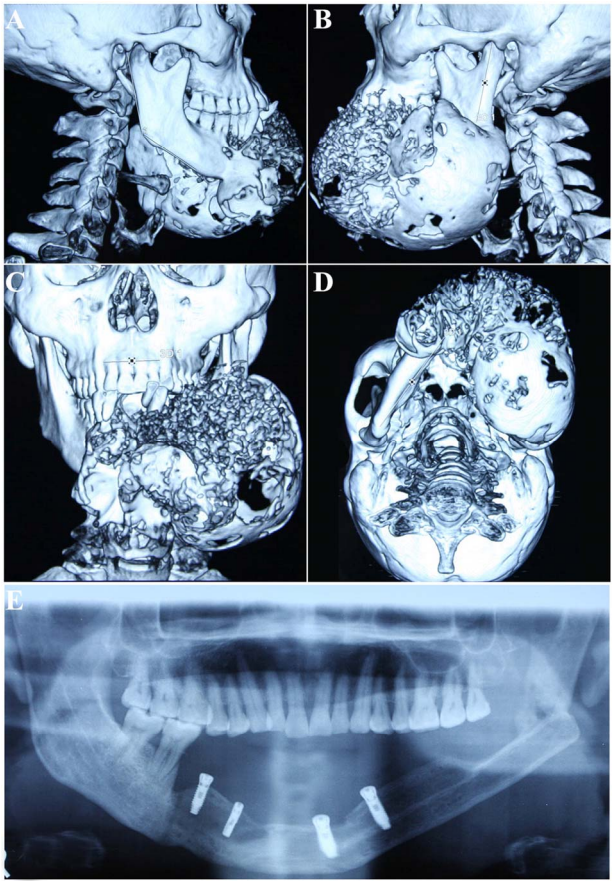

Figure 5. Radiological findings of Case 2. Preoperative CT scanning with 3 dimension reconstruction (A)-(D) demonstrated a great multicystic region mainly in the left mandibular area including the body and ramus. The panoramic radiograph (E) showed good bone healing and placement of dental implants in the distracted fibula graft.

Figure 6. Intra-operative photographs of Case 2. A showed the cutting and shaping of the fibula. B showed good contour and integration of grafted fibula before placement of distractor. C showed inverted trapezoidal osteotomy in the front jaw and 2 implanted distracters.

the steady levels of bone around the fixtures could be-seen (**Figure 6(B)**). In order to prevent interference with each other during the distraction, inverted trapezoidal osteotomy was made in the front jaw, and then 2 distractors were implanted (**Figure 6(C)**). Rod of the distractors pointed to the function surface of maxillary teeth.

3.4. Step 4 Distraction Osteogenesis

After a latency period of 7 days, the distractor was activated 3 times daily (1.0 mm per day). The duration of distraction lasted for 10 - 15 days until the height of front jaw reaching 2.5 - 3 cm.

3.5. Step 5 Distractor Removal

After a consolidation period of 3 months, the patient underwent the fifth surgical stage, removal of the distractor through the submandibular incision. New bone with sufficient volume and density, which was depicted radiographically was confirmed clinically during the process of removing distractors.

3.6. Step 6 Placement of Endosseous Dental Implants

Dental rehabilitation plan was conducted 3 months after the distractor removal. Following clinical and radiological evaluations, the patients underwent vestibular plasty and placement of endosseous dental implants. Two and four OSSTEM GS Ⅲ implants with a diameter of 4.5 mm and a length of 11.5mm (Manufacturer OSSTEM IMPLANT Co., Ltd.#507-8, Geoje 3-dong, Yeonje-gu, Busan, Korea) were placed into the reconstructed area for Case 1 and 2, respectively, according to the manufacturer's instructions. Locations of the implants were determined by temporary denture, while long axis of the implants point to the function surface of maxillary teeth.

3.7. Step 7 Prosthodontic Rehabilitation

The final prosthetic rehabilitation was performed 3 months after the placement of implant. Ball attachment was chose for further prosthodontic rehabilitation.

At the end of the sequential treatment, chewing and speech functions of the patients were restored, without surgical complications. The esthetic and functional outcomes restituted a satisfactory quality of life to the patients. The four-year follow-up proved the optimal integration of grafted tissue and the steady levels of bone around the fixtures. Peri-implant soft tissues are healthy.

4. DISCUSSION

Ameloblastoma is the most common tumor of odontogenic origin. Clinically, it is a painless, slow-growing and persistent lesion, but behaves as an invasive and recurring tumor in spite of its benign histological nature [1,2,10]. If not treated, ameloblastoma can gain an enormous size and cause severe facial disfigurement and functional impairment [9]. Both the patients reported here delayed treatment because of economic reasons; as a result, the tumor grew to such an extent and impacted their life seriously.

Recurrence is another terrible feature of ameloblastoma. It has been reported that more than 50% of recurrence appears within the first 5 years after primary sur-

gery [2], and the risk of recurrence mainly depends on the method of treatment and the type and size of the tumor [1,10]. From a surgical standpoint of view, it has been reported that the recurrence rate following a simple curettage is much higher than radical resection, 75% and 20%, respectively [1]. So radical resection was applied to both the two patients reported in this article. And after a 4-year follow-up, no evidence of tumor recurrence was found in the patients. However, radical surgical ablation procedures will inevitably result in large tissue defects, which lead to severe aesthetic and functional sequelae, with a significant loss in the quality of life unless it is reconstructed successfully [9]. The patient's postoperative quality of life largely depends on the quality of the mandibular reconstruction. So how to restore the patient's appearance and mandible function is the key to the treatment of huge ameloblastoma.

In recent years, vascularized bone grafts are widely considered as a reliable technique for reconstructing segmental mandible defects. Because in comparison to non-vascularized free bone grafts, microsurgical transfers of free bone grafts can reconstruct mandible defects with an immediate source of blood supply to the graft [2]. Fibula, ilium, scapula, and radius are the 4 commonly used osteocutaneous flaps for mandibular reconstruction [9]. Among these flaps, fibula and iliac crest are the most commonly used free flaps. In this series, we chose fibula flap rather than iliac crest, giving full consideration to fibula's unique advantages, including providing sufficient bone segment for any length of mandibular defect, possibility of multiple osteotomies because of both endosteal and segmental blood supply, re-establishing the contour of the mandible, correction of intermaxillary relation, less resorption, suitable for insertion of dental implants because of its proper bone thickness and bi-cortical structure, flap dissection under tourniquet with minimal blood loss, and a long pedicle up to 8 cm in length [2,9]. Furthermore, the skin flap of the fibula is thin and pliable which is suitable for use as oral lining [9]. And the four-year follow-up of the patients proved that continuity and contour of the mandible was reconstructed well. However, we also have to admit that limited bone height comparing with a dentate mandible is the main disadvantage of this flap [9,11]. The loss of vertical bone height results in an unfavorable crown-root ratio when dental implants are planned for occlusal rehabilitation. In this series, we addressed this problem via vertical DO on the fibula graft.

In the last decade, DO become a popular modality in correcting craniomaxillofacial bone malformations [12], especially in managing the mandibular hypoplasia, mandibular defect and loss of alveolar ridge height [2,13]. As far as we know, only a few cases of vertical DO of a fibula flap have been reported in the literature [14-21]. However, the advantages of DO, such as distraction of soft tissue along with lengthening the bone, the predictable outcome, the simplicity of the procedure, the lower postoperative morbidity without the necessity for bone grafts or donor sites, determines vertical DO is an excellent and reliable method to increase fibular bone height according to the individual local needs [13-21]. So we chose vertical DO on the fibula graft to increase the ridge volume before placement of dental implants. And the results showed that the quality of the neogenerated bone is excellent with adequate characteristics for implant osseointegration, fully ensuring the following dental implantation and prosthodontic rehabilitation. What is worth mentioning is the need to wait 6 months after fibula transfer before performing DO. Because during mandibular reconstruction, good contour of the fibula usually required multiple osteotomies, which interrupted the medullary vessel [14,19]. Long enough interval can ensure complete bone regeneration.

5. CONCLUSION

This report presents two patients affected by extensive mandibular ameloblastoma. Both the patients underwent radical resection of tumor and simultaneous mandibular reconstruction by fibula free flap. Vertical DO on the fibula graft was applied in order to obtain adequate bone height and to realize placement of dental implants. After osseointegration, the patient was rehabilitated with ball attachment denture. The entire course of treatment takes only about 15 months. After the four-year follow-up, neither soft tissue related, nor hard tissue, nor implant related problems were observed. Satisfactory facial symmetry, chewing and speech functions of the patient were restored. So this sequential treatment for extensive mandibular ameloblastoma can obtain an excellent effect by the shortest time and the lowest economical cost. Furthermore, the series also can be used to reconstruct giant mandibular defects caused by different reasons.

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