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Case Report

The Role of Free Fibula Flap in the Reconstruction of a Mandibular Ameloblastoma: A Case Report

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Abstract

Introduction: The ameloblastoma is a rare odontogenic tumor of the oral cavity. It affects more the mandible than the maxilla, and has a predilection for the posterior region. Although this tumor is benign, its behavior is locally aggressive and requires the most often surgical resection margin.

Case Report: A young woman aged 28, has consulted the Oral Surgery Department of Rabat, complaining of a right mandibular swelling, extended from the 46 to the right mandibular ramus, lasting for eight months. The intraoral examination revealed a swelling covered of a mucosa of normal appearance. Panoramic radiograph revealed the presence of a multi-geodic lesion at the right hemi-mandible. A biopsy was performed at the level of the lesion and concluded an ameloblastoma. The patient was subsequently referred to the Maxillofacial Surgery Service of the Hospital of Specialties of Rabat. Two teams, one of maxillofacial surgery and another one for vascular surgery, collaborated to perform a hemi-mandibulectomy with a free fibula flap graft.

Conclusion : The free fibula osteocutaneous flap is the most versatile and reliable option for microsurgical reconstruction of large mandibular defects.

Keywords: Free Fibula Flap ; Ameloblastoma ; Mandibular Reconstruction

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Introduction

Ameloblastoma was firstly described by Cusack in 1827. It is the most common benign odontogenic tumor and represents 13–54% of all benign and malignant tumours of the jaw1. There is no distinct gender predilection and most cases are diagnosed in the third to fifth decades of life. While rare in children younger than age 10 years, it is relatively uncommon in the 10 to 19 year old group. The ratio of mandibular to maxillary ameloblastomas is 5:1 and usually involves the molar-ascending ramus area. Ameloblatomas may be classified into two groups- benign and malignant. Malignant variants are extremely rare and constitute less than 1% of all ameloblastomas. Benign ameloblastomas are grouped into three different patterns solid/multicystic, unicystic and extraosseous/peripheral. Solid/multicystic ameloblastomas are most common, with a slow but infiltrative growth pattern. They are locally aggressive and occur mainly in adults. They present as multilocular radiolucent lesions often described as ‘honey- combed’ or ‘soap-bubble’ appearance. Frequently, an unerupted tooth is associated with the lesion.

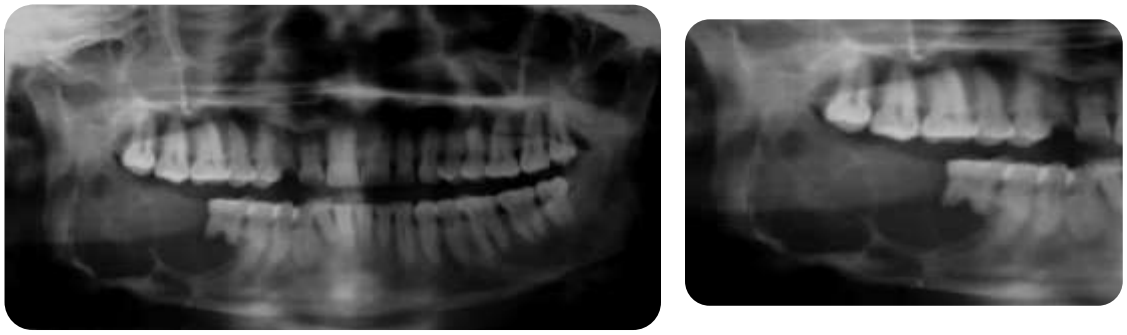
Case Report

A 28 years old female reported to the department of Oral Surgery of the Center of Consultation of Dental Treatment of Rabat, complaining from pain in the right mandibular region lasting for about 8 months. The general history of our patient was unremarkable.

The extraoral Examination showed an asymetry as well as the presence of a swelling in the right posterior mandibular region (Fig. 1). There were no lymph nodes noted.

Fig. 1 Extraoral view : Facial asymetry with the presence of a mandibular right swelling in the posterior region extended from 45 to the ramus.

The X ray findings revealed the presence of multilocular radiolucent lesions in the right posterior mandibular region, extended to the right ramus (Fig. 2).



(A) **(B)**

Fig. 2 Orthopantomogram incidence showing multilocular radiolucent lesions in the right posterior mandibular region, extended to the right ramus.

Based on the clinical examination, the radiological findings and the localization of the lesion (mandibular angle and ramus), we could evoke many diagnostics as ameloblastoma, keratocystic odontogenic tumor, odontogenic myxoma, ameloblastic fibroma, or even a central giant cell granuloma. To establish a final diagnosis, we did perform a biopsy which concluded an ameloblastoma.

The patient was then referred to the maxillofacial department in the multispecialities hospital of Rabat to perform the excision of the tumor. A second team of vascular surgery collaborated and could successfully perform the fibular sample to restore the defect caused by the ameloblastoma. After the surgical excision of the tumor by hemomandibulectomy (Fig. 3), the specimen was sent for histopathology study zhich revealed the presence of a multicystic follicular ameloblastoma. (Fig. 4). Then vascular surgeons sampled the vascularized fibula graft and carefully placed it in the defect area, while the maxillofacial surgeons maintained this fibula graft with the ostesynthesis plates (Figs. 5,6,7). The patient had no post-operative complications and couldn't come for follow-up due to his unavailability.

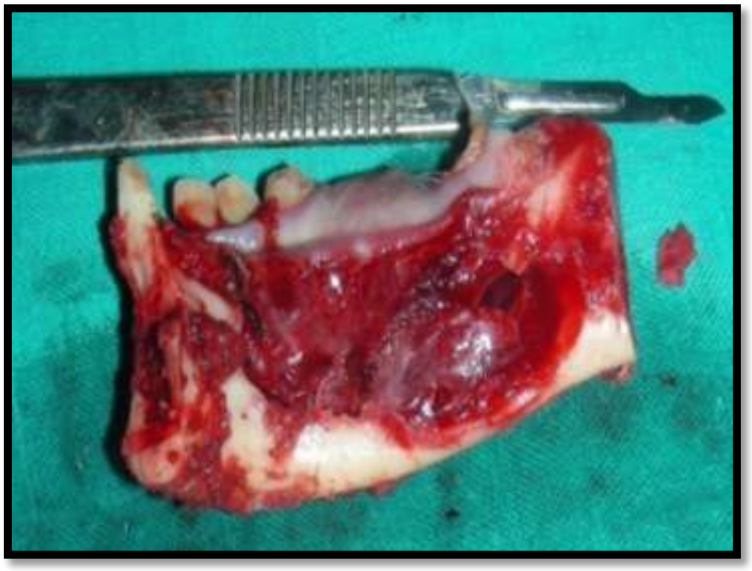


Fig. 3 The tumor's specimen after hemimandibulectomy

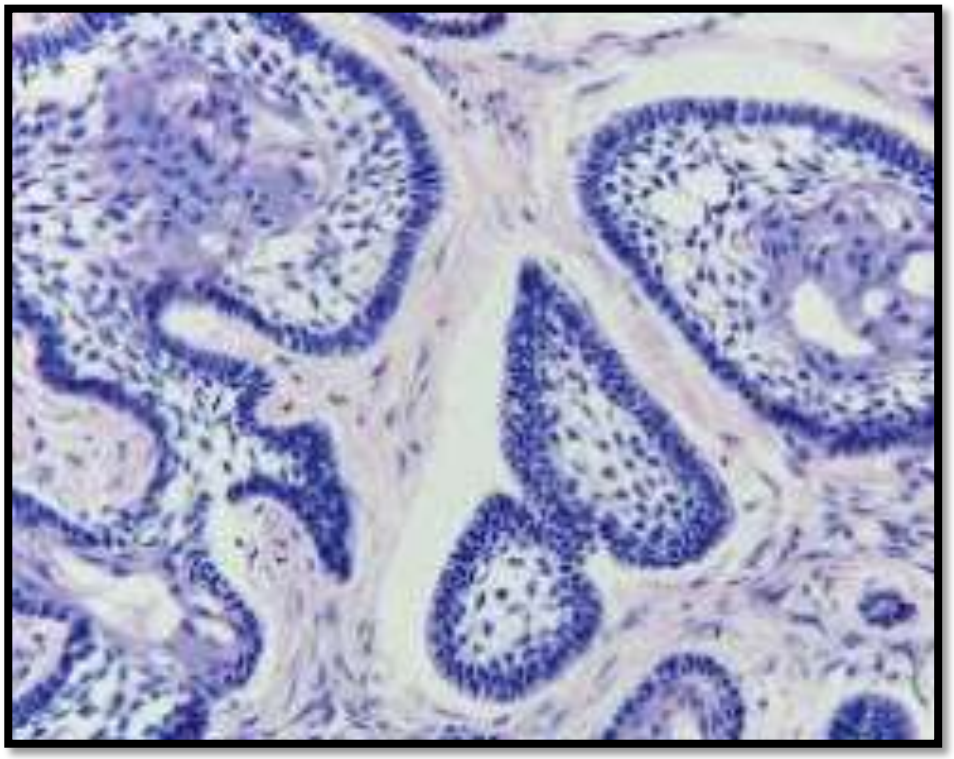


Fig. 4 Histopathological pattern revealing a follicular multicystic ameloblastoma.

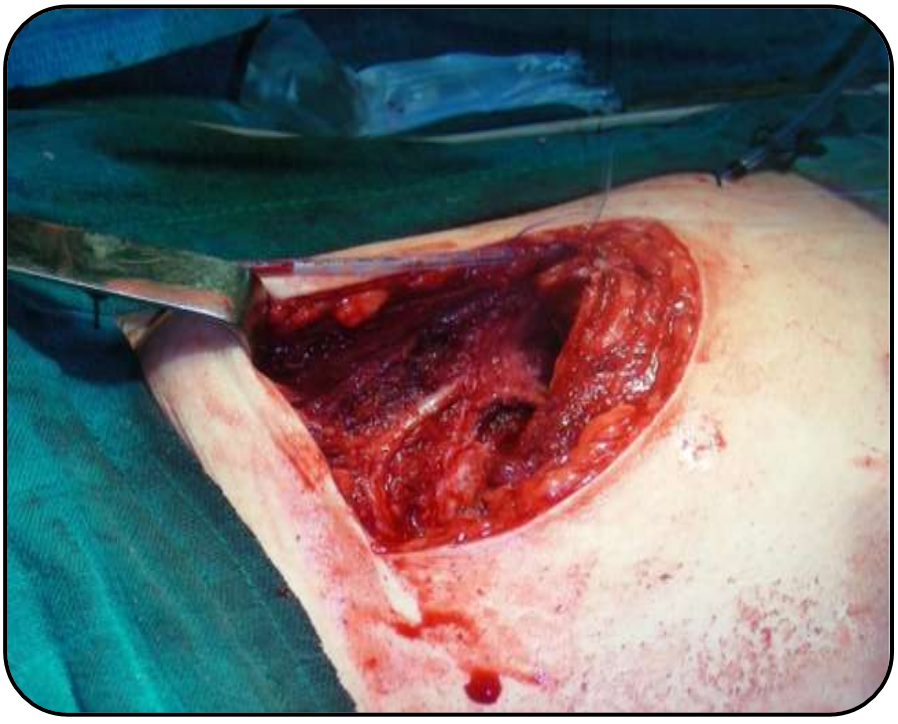


Fig. 5 The emergence of the Fibula bone during the sampling by the Vascular Surgery team.

Fig. 6 The free Fibula Graft specimen

Fig. 7 The free fibula flap repositionning in the mandibular defect zone and maintained with the ostesynthesis plates.

Discussion

Ameloblastoma is the most common benign odontogenic tumor and represents 13–54% of all benign and malignant tumours of the jaw. The World Health Organization **(WHO)** defined ameloblastoma as a locally-invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma. Its behaviour has been described as being benign, but locally aggressive [1].

There is no distinct gender predilection in ameloblastomas and most cases are diagnosed in the third to fifth decades of life. While rare in children younger than age 10 years, it is relatively uncommon in the 10 to 19 year old group. The ratio of mandibular to maxillary ameloblastomas is 5:1 and usually involves the molar-ascending ramus area [1,2].

Ameloblatomas may be classified into two groups benign and malignant. Malignant variants are extremely rare and constitute less than 1% of all ameloblastomas. Benign ameloblastomas are grouped into three different patterns — solid/multicystic, unicystic and extraosseous/
peripheral. Solid/multicystic ameloblastomas are most common, with a slow but infiltrative growth pattern. They are locally aggressive and occur mainly in adults. They present as multilocular radiolucent lesions often described as ‘honey-combed’ or ‘soap-bubble’ appearance. Frequently, an unerupted tooth is associated with the lesion [3].

Up to 80 % of ameloblastoma cases occur in the mandible, with a predilection for the posterior mandibular region. Rare cases have been reported as primary to the sinonasal cavities. Ameloblastoma can be associated with unerupted third molar teeth, particularly in the unicystic type. Desmoplastic ameloblastomas often occur in the anterior or premolar regions of the mandible or maxilla. Ameloblastic carcinomas also favor the mandible (*2/3) over the maxilla. Maxillary ameloblastomas also mostly occur in the posterior molar region [2,3].

Why radical treatment for ameloblastomas ?

Surgery is the standard treatment for ameloblastomas. Historically, the extent of resection has been controversial, comprising of two surgical options: ‘‘conservative’’vs.‘‘radical ». The former involves enucleation/curettage of the bony cavity, while the latter involves a radical operation with appropriate margins. Advantages of enucleation include the fact that it is an outpatient procedure able to be performed by many different service providers (Oral Surgeons and ENT), since it requires no reconstruction. Historical data on simple enucleation demonstrates recurrence rates 60–90 %. However, this treatment modality is currently believed to play no role in the management of multicystic ameloblastomas [4].

The ‘‘radical’’ surgical option is the current standard of care for ameloblastoma and includes en bloc resection with 1–2 cm bone margins and immediate bone reconstruction to help with speech and swallowing. The bony margin is defined as the distance away from the radiographic margin predicted to be disease free and oncologically safe to perform osteotomies. Data from 82 ameloblastoma specimens showed microscopic tumor extension 2–8 mm (mean of 4.5 mm) beyond the radiographic boundaries of the tumor. Hence recommended bone margins are 1–1.5 cm for unicystic and 1.5–2 cm for solid/multicystic histological types, and provides increased cure rates.

For all other WHO-classified mandibular ameloblastomas, a segmental resection which includes at least one adjacent uninvolved anatomic barrier for proper margins is advocated. The healthy mucosa overlying the cortical perforation is often removed as a margin. Segmental resection of the

mandible results in discontinuity of the jaw, which is stabilized to its previous position by titanium reconstruction plates to ensure proper occlusion [5,6].

What graft to choose ?

Although free nonvascularized grafts as first described by Lexerl continue to be used, the last decade has seen substantial advancement in the restoration of mandibular continuity defects largely through the introduciton of vascularized bone grafts. Cases involving non or poorly vascularized graft beds, simultaneous bone and soft tissue replacement, or extensive bone defects call for the use of such grafts. The vascularized rib was one of the first bone grafts to be used in mandibular reconstruction. Donor sites for microanastomized grafts in current use include the iliac crest, the scapula, the radius, and the fibula. These grafts have their specific drawbacks [7-9].

The radius, for instance, has a limited length, and the scapula has limited width. The limitations in the width of the radius and fibula result in height deficiencies of the reconstructed segment when these transplants are used in dentate or nonatrophied edentulous mandibles." During extensive reconstruction, it is often desirable that one surgical team harvests the graft as the other prepares the donor site. Use of the scapula in mandibular reconstruction precludes this possibility. The iliac crest transplantation however, demands longer periods of patient immobilization, introduces contour defects in the iliac bone, and can lead to severe donor site morbidity, such as damage to the femoral nerve, pelvis fractures, abdominal herniation, peritonitis, **« bone resorption »** and protracted gait pain [7,8].

The free vascularized fibula graft was first reported in 1975 by Taylor et al. They successfully used the graft to bridge a large posttraumatic defect of the contralateral tibia. In the ensuing years, the vascularized fibula transfer gained in popularity, and its indications were extended. Fibula grafts were not used in oral and maxillofacial reconstruction until the beginning of the 1980s. They have nevertheless come into wide use since their introduction [9,10].

The Vascularization of the fibula

One way of avoiding a large vertical dimension of the prosthetic suprastructure is the use of a long fibular graft that is halved and folded onto itself to increase the height of the neomandible. The possibility of segmenting and folding the graft onto itself is facilitated by the unique blood supply to the fibula shaft. The peroneal artery courses with the corresponding two peroneal veins parallel to the long axis of the fibula. These peroneal vessels supply a centromedullary and a periosteal vessel network responsible for the blood supply to the fibula shaft. The centromedullary flow is effected largely through the nutricium artery, which normally enters the bone via the nutricium foramen before dividing into an ascending and a descending branch. The foramen is located within the cranial half of the middle third of the fibula shaft. Perpendicular osteotomies of the fibula to divide it into several segments do not compromise the blood supply to the segments [9-11]. Whereas the segment containing the foramen nutricium is supplied by both the medullary and the periosteal networks, the blood supply to the proximal and distal segments is maintained by the periosteal network alone. Moreover, it increases the donor site morbidity significantly. The use of implant restorations based on free fibula flaps was first reported in the early 1990s. Since then, several studies have evaluated the different prosthetic options available and the long-term results. Good long-term results have been reported using dental implants in fibula flaps. For example, Ghara et al reported 4 failures of 121 implants placed in 30 patients who had

undergone a free fibula flap reconstruction. They highlighted the great improvement in the quality of life of their patients [12,13].

When the free fibula flap is compared with the scapular or radial osteocutaneous free flaps, one of its main advantages is the better bone quality. It is perfectly able to support an implant prosthetic rehabilitation. Although the radial forearm flap also has many desirable characteristics, it is associated with an unacceptably high rate of fracture in the residual radius. The greatest disadvantage of the scapular flap is that it takes much longer than a fibula flap. In addition, significantly less bone is available than with a fibula flap. Therefore, although large series have reported good results for mandible reconstruction with the radial forearm osteofasciocutaneous flap or scapular osteocutaneous flap, neither is currently considered a first reconstruction option. A problem with the fibula graft in this indication is the large difference in height between the reconstructed and the intact mandibular segments when the mandible is dentate or nonatrophic [14,15]. The lack of adequate alveolar ridge after free fibula bone transfer can make prosthodontic rehabilitation of these patients challenging. The 'double-barreling' of the fibula to create equal struts is a useful modification with good aesthetic and functional outcomes. Partially double-barreled grafts, as needed for aesthetic improvements, have also been reported, and a commonly used modification [16,17]. The 'double-barreling' of the fibula enables a suitable height of the alveolar ridge, but also an immediate osseointegrated dental implantation, obtaining better height results and lower complication rates compared to vertical distraction devices [18-20].

Conclusion

In our experience, the free fibula osteocutaneous flap is the most versatile and reliable option for microsurgical reconstruction of large mandibular defects. It provides a large quantity of bone, which can be shaped easily to adapt to the remaining mandible passively. The bone height is suitable for an implant-based prosthetic restoration. Preoperative mapping of the cutaneous perforators improves the versatility of the flap design and decreases the donor site morbidity [20].

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