

## Kintsugi flap in the management of giant mandibular ameloblastoma: A case report

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### ABSTRACT

Neglected ameloblastomas may present with massive swelling of the jaw. Reconstruction of intraoral mucosal and floor of the mouth defects following surgical resection of the tumors is always a challenge. However, it is not possible for all patients due to various reasons which include a medical status that precludes an extended operative procedure, history of coagulopathy, connective tissue disorders, vasculitis, peripheral vascular disease, venous insufficiency, and the second surgical site. Reconstruction of the mandible with titanium plate is commonly practiced for those patients. We report the case of giant ameloblastoma measuring approximately 14 cm × 13 cm × 10 cm, involving the right side of the mandible crossing midline up to the left body of the mandible. The tumor was excised and the mandibular defect was reconstructed with titanium plate fixed. The purpose of this article is to propose a possibility of using the redundant tumor-free skin and subcutaneous tissue as a local cutaneous flap for the reconstruction of the floor of the mouth and as a soft tissue draping for the titanium plate. The technique has the advantage of avoiding a second surgical site and has no donor site morbidity. Being done as a single-stage procedure, it eliminates the need for revision of the flap, thereby avoiding multiple surgeries.

**Key words:** Giant ameloblastoma, Intraoral reconstruction, Kintsugi flap, Local cutaneous flap

**A**meloblastoma is a benign locally aggressive tumor arising from the odontogenic ectoderm. It accounts for 1% of all oral tumors and for 9–11% for all odontogenic tumors [1]. In 80% of the cases, it is localized in the mandible, and in 20% cases, it affects the maxilla [2]. Authors have expressed different opinions on the sex and age distribution. Male predominance has been reported in Africa, North America, and Asia. The peak incidence of ameloblastoma, worldwide, is in the third decade. In Asia, the incidence is between the third and sixth decades. In Europe and North America, ameloblastoma mostly occurred at an older age (the fifth and sixth decades) while in Africa and South America, ameloblastoma mostly occurred at a younger age (the third decade) [3]. On late presentation, ameloblastoma may become enormous and cause gross facial disfigurement that poses considerable problems in management [4].

The treatment of choice at this stage is resection of the involved mandible and musculature of the floor of the mouth. When alloplastic (titanium plate) reconstruction of the mandible was done, replacing an adequate amount of intraoral soft tissue for the defect in the floor of the mouth is significant to the overall success of reconstruction. Conventionally, local flaps such as nasolabial flap, tongue flap, submental flap, and facial artery musculocutaneous flap were used for the reconstruction of intraoral defects. The limitations of these flaps include small size which can cover only small-to-medium defects and thinness of flap is not suitable for providing bulk [5].

We propose the technique of using the redundant tumor-free skin and subcutaneous tissue as a local cutaneous flap for the reconstruction of intraoral defects in the case of giant ameloblastoma. The advantages of this technique are that the flap harvest is easy, it can be done in a single-stage procedure, and most of all, there is no donor site morbidity.

A 51-year-old woman reported to our department with a huge painless swelling on the right side of the mandible crossing the midline that had gradually increased over the past 25 years to attain the present size (Fig. 1). The patient had difficulty in eating and swallowing. During the initial phase of the disease, the patient consulted various dentists and maxillofacial surgeon, and they have advised surgical excision. However, the patient was very apprehensive for the surgery and kept denying the procedure. Over time, the swelling enlarged, causing severe facial disfigurement. This had a psychological impact and the patient isolated herself from society other than her own family.

On general examination, the patient was conscious, well oriented to time place, and person with no signs of pallor, icterus, cyanosis, clubbing, pedal edema, and normal gait. The patient vitals were recorded which was within the normal range and the patient had no co-morbidities. On local examination, the patient had gross facial asymmetry affecting the right side of the face. The swelling was approximately 15 cm × 13 cm extending superiorly up to the infraorbital rim, inferiorly causing

bowing of the lower border of the mandible, medially crossing the midline, and laterally up to the tragus of the ear. The skin over the swelling was stretched with no rise in temperature and no signs of infection.

Computed tomogram scan revealed an expansible lesion containing both solid and cystic components, predominantly cystic measuring approximately 14 cm × 13 cm × 10 cm encompassing the entire right side of the mandible also extending up to the left body of the mandible (Fig. 2). An intraoral biopsy confirmed follicular ameloblastoma.

The tumor was excised en-bloc with supra periosteal segmental mandibulectomy through a modified “visor” neck incision under general anesthesia. As the patient was not willing for a secondary surgical site (osseocutaneous fibular free flap), the resulting mandibular defect was reconstructed with titanium plate fixed to the remaining mandible on the left side. The floor of the mouth was reconstructed with a transpositioned local cutaneous flap using the tumor-free redundant tissue.

Preoperatively, vertical and horizontal measurements were made on the normal as well as the affected side. On the normal side, the vertical measurement was made from the outer canthus of the eye to the lower border of the mandible; the horizontal measurement was

made from the tragus of the ear to the corner of the lip. The approximate measurement was 6.5 cm × 11.5 cm. The same measurements were repeated on the affected side, which was 15 cm × 16 cm. The excess skin on the affected side was 8.5 cm × 4.5 cm. A modified “Visor” incision was made starting from the first neck crease on the contralateral side, crossing the midline and gently curving over the pre-auricular skin on the tumor side.

The tumor was excised en-bloc with a suprapariosteal segmental mandibulectomy (from the condyle to the left body of the mandible region). Care was taken not to create any buttonhole in the flap on retraction. The resulting mandibular defect was reconstructed with titanium plate fixed to the existing mandible (Fig. 3). Primary closure was not possible for the intraoral defect measuring approximately 10 cm × 3 cm in the greatest dimension.

A cutaneous flap measuring 12 cm × 3.5 cm was raised from the redundant tissue on the tumor side, maintaining a broad base on the non-tumor side (Fig. 4a). Near the junction of the pivot point of the flap, the skin was de-epithelialized and sutured to the skin along the incision to avoid flap revision and second surgery. The flap was transpositioned intraorally to cover the titanium plate and to reconstruct the floor of the mouth (Fig. 4b). The primary incision margins were approximated and closed without tension.

During the post-operative period, the patient was on nasogastric tube feeding and excellent oral care was maintained for 10 days. The entire post-operative phase was uneventful. At 6 months follow-up period, there was complete healing of the flap without any exposure to the reconstruction plate. There was also no evidence of oro-cervical fistula (Fig. 5).



Figure 1: Pre-operative image (published with the patient’s consent)

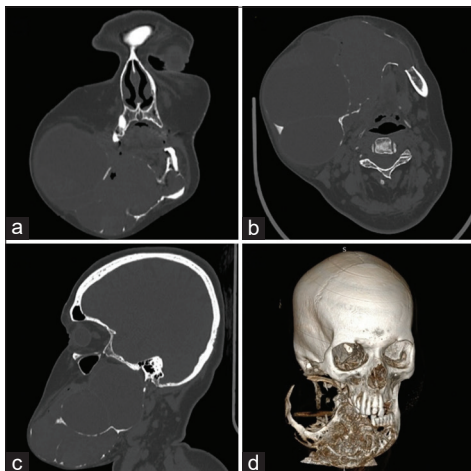


Figure 2: Pre-operative computed tomography (a) coronal view, (b) axial view, (c) sagittal view, and (d) three dimensional reconstruction view



Figure 3: Mandibular reconstruction with a titanium plate

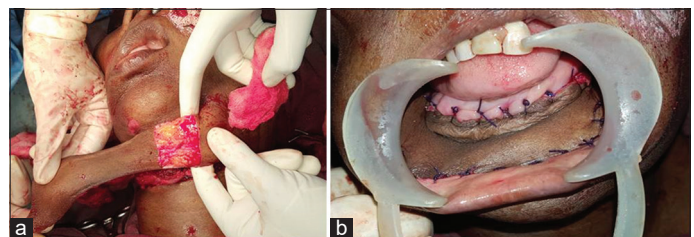


Figure 4: (a) Flap raised from expanded skin and subcutaneous tissue; (b) the flap transpositioned intraorally to drape the titanium plate and to reconstruct the floor of the mouth

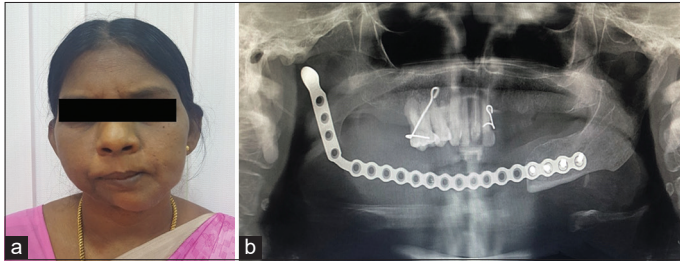


Figure 5: Six months (a) post-operative image and (b) post-operative orthopantomogram

## DISCUSSION

The term giant mandibular ameloblastoma is used when (a) the size of the tumor is more than 5 cm in length, (b) thinning of the inferior and or posterior mandibular borders, and (c) considerable jaw expansion [6]. Giant ameloblastomas are commonly treated with a radical approach. The method of mandibular reconstruction after segmental mandibulectomy, such as vascularized osseous-cutaneous flap transfer, a combination of reconstruction plate with or without soft tissue transfer is chosen based on various factors such as the patient's general condition, the extent of the resection, patient's age, consent of the patient for the second surgical site, and financial status.

The use of reconstruction plates for mandibular reconstruction is often criticized because reported rates of post-operative plate exposure have been extremely high such as 46.15% reported by Wei *et al.* [7]. Mariani *et al.* (2006) have reported a significant rate of plate exposure when the resection includes the central segment of the mandible [8]. The other factors attributing to plate exposure are (a) persistence of dead space below the plate, (b) the soft tissue over the plate surface, (c) imbalance between the soft tissue on the plate surface and the form of the plate, (d) necrosis of soft tissue surrounding a plate, (e) allergy to metal, (f) contouring of the plate, and (f) effects of radiation therapy [9]. However, Chepeha *et al.* showed that the rate of such complications could be reduced by over-reconstruction of the soft-tissue defect to support the plate medially which lowers the risk of plate extrusion [10].

Transposition flaps are usually designed as random cutaneous flaps but may occasionally be axial or compound. Although most flaps on the face have a random blood supply, the abundant vascularity of the skin of the face enables the successful transfer of flaps that exceed the 3:1 ratio of length to width [11].

In our case, there was no evidence of distant metastases which is more common in large, long-standing, or those treated conservatively with repeated surgeries [12]. Irrespective of its large size, the tumor did not penetrate neither to the periosteum nor the skin. We considered the expansion of tissue due to the underlying tumor like the soft tissue expansion accomplished by tissue expander used in the maxillofacial region. The possible complication with this surgical technique can be plate exposure, infection, and necrosis of flap. Our patient had none of the complications and the recovery was uneventful.

Studies of the flap survival, which is dependent on the skin vascularity, reveal that flaps raised from expanded tissue have survival rated comparable to those of delayed flaps. Cherry *et al.*

demonstrated that flaps raised from expanded tissue have a 117% increase in survival rate compared with random pattern flaps raised on non-expanded skin [13]. Kintsugi means golden joinery in Japanese. As a philosophy, if anything has been damaged, then it has more of a history and should, therefore, be highlighted, rather than discarded. On that note, we would be happy to call this flap as “Kintsugi flap.”

## CONCLUSION

The flap which we used with the expanded redundant tissue served as an excellent local flap for intraoral reconstruction. It can be used in a single-stage procedure that has the advantages of high vascularity, less technique sensitive, economically feasible and has no donor site morbidity.

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