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Case Report

Management of Posterior Maxillary Ameloblastoma with Scapular Tip Free Flap

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Abstract

Maxillary ameloblastomas are rare odontogenic epithelial tumors predominantly located in the posterior segment of the maxilla. Due to their anatomical confinement and difficulties for successful resection, these tumors show high risk of postoperative complications and recurrence. The reconstruction with vascularized bone grafts appears to be a reasonable option for optimal cosmetic and functional results. We present a case of maxillary ameloblastoma located at molar region treated with extensive resection and scapular tip free flap.

Keywords: Maxillary ameloblastoma; Maxillectomy; Radical resection; Spular tip free flap

Introduction

Ameloblastomas are benign, but locally invasive tumors originated from remnants of dental lamina, enamel organ and odontogenic epithelium [1]. They are responsible for 1% of all tumors and cysts of the jaws and 10% of odontogenic tumors [1-3]. They are mainly seen in adults with male predilection and more frequently located in the mandible [4]. Ameloblastomas are classified into four different categories namely multicystic, unicystic, desmoplastic and peripheral [5]. Even though conservative approaches have been described (i.e., enucleation, curettage), extensive surgical excision seems to be the only modality of treatment that secure reasonable curative rate [2,6]. Ameloblastomas can experience locoregional recurrence, develop distant metastasis and even malignant transformation [7-9].

Maxillary ameloblastomas are mostly located in the molar region and frequently progress into adjacent tissues and anatomical spaces [10,11]. The diagnosis is established with the clinical examination, radiologic appearance and histological findings. Maxillary ameloblastomas may exhibit minimal or no symptoms at the time of the diagnosis [12]. However, they are able to develop toothache, loose teeth, intraoral ulceration, edema or nasal obstruction, nasal epistaxis and visual disturbances. In general, symptoms appear when the lesion has already advanced beyond the maxillary bony boundaries [13]. In imaging tests, maxillary ameloblastomas display unilocular or multilocular radiolucency features associated with destruction of cortical bony walls [14]. They can present different histopathologic patterns, but follicular is the more frequent variant [15].

Case Report

A 54-year-old African American male who presented to our clinic after dental extraction of impacted tooth #1 and biopsy that revealed right posterior maxillary ameloblastoma. Patient denied weight loss, pain, loose teeth, dysphagia, sensory deficit, facial edema, sinus congestion or discharge. His past medical history included controlled hypertension and abdominal surgeries (appendectomy, small bowel obstruction). No history of tobacco or alcohol consumption.

On physical examination, his neck appeared symmetric with normal range of motion, soft, non-tender and with no palpable lymphadenopathies. Normal temporomandibular joints assessment and good dentition with stable and reproducible occlusion. Maximum Interincisal Opening (MIO) of 40-mm. Normal intraoral

soft tissue appearance with well healed socket from previous extraction and no obvious exophytic mass or expansion of bone in maxilla. Panoramic dental radiograph and CT neck demonstrated a 2.8 x 2.4 x 1.5 cm expansile lesion centered in the posterolateral aspect of the right alveolar maxilla (Figure 1). Other findings were unremarkable except impacted tooth #16 and unerupted tooth #32. The chest imaging assessment was normal except for thoracic scoliosis. CTA of lower extremities showed that posterior tibial arteries were diffusely small in caliber and peroneal arteries provided blood supply to plantar regions. CTA of chest revealed a favorable vascular anatomy for a right scapula flap. The presence of circumflex scapular artery and angular branch of thoracodorsal artery were confirmed with handheld Doppler. Based on patient's body composition (BMI 22 kg/m^2) and Doppler findings, we decided to perform an ipsilateral Scapular Tip Free Flap (STFF). Prior to the surgery, a virtual surgical planning session was carried out to design a pre-bent plate and surgical cutting guides for both maxillary and scapular resection (Figure 2(A-B)).

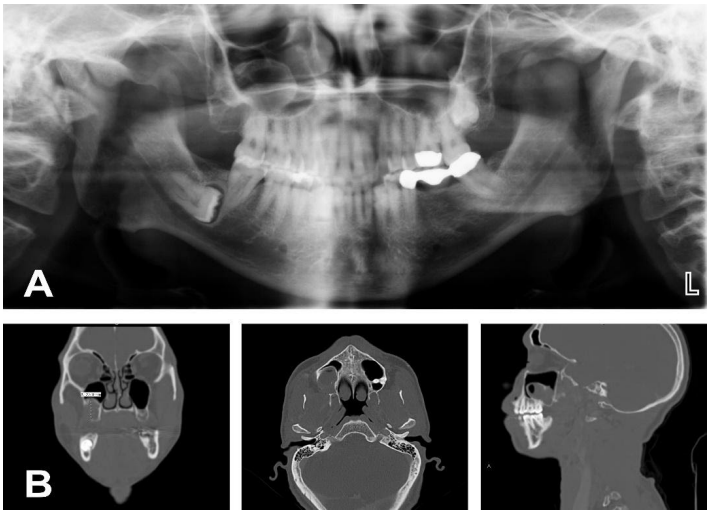

Figure 1: 54-year-old African American male with ameloblastoma of right maxilla. Patient was asymptomatic until the tumor was discovered during a tooth extraction. Past medical history of controlled high blood pressure and permanent use of 2 antihypertensive medications. No consumption of tobacco or alcohol. BMI of 22 kg/m^2. Panoramic dental radiograph **(A)** and CT scan **(B)** demonstrated a 2.8 x 2.4 x 1.5 cm ameloblastoma centered in the posterolateral aspect of the right alveolar maxilla.

Patient underwent general anesthesia with endotracheal intubation through the left nostril. To maintain the same position during the entire procedure, the patient was placed in a modified lateral decubitus at 30 degrees of right hip elevation and 45 degrees of chest rotation (Figure 2C). This position was achieved with foam padding and suction beanbag. The opposite upper extremity was cradled without pressure or distraction on the shoulder. In addition, adequate padding was applied around knees, ankles and feet. During the surgery, we modified the lateral inclination of the operating table every 1-2 hours to change the points of maximum pressure on the skin. The right upper extremity was prepped and covered with sterile sleeve to facilitate its movements and the dissection within the surgical field. A sterile handheld Doppler was utilized to localize and define trajectory of vessels of interest, and subsequently, confirm the integrity of the resulting flap pedicle. An incision on the right neck was performed (2-cm below the mandibular margin) by the ablative team to expose neck vessels in preparation for microanastomoses. Using intraoral approach, and after appropriate elevation of soft tissues, a right partial maxillectomy was initiated with reciprocating saw over prefabricated cutting guides. Then, through a series of straight and curved osteotomes, the specimen was finally freed and passed off to the back table.

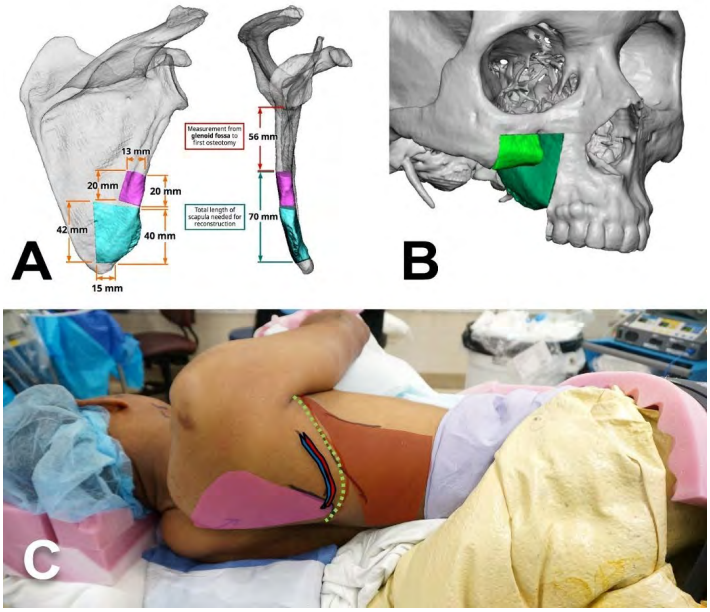

Figure 2 (A-C): Virtual surgical planning. **(A)** vascularized (light blue area) and non-vascularized (magenta area) bone grafts from right scapula. **(B)** Tentative right partial maxillectomy followed by placement of vascularized (dark green) and non-vascularized (light green) bone grafts on surgical defect. **(C)** Position of the patient on operating table. Dotted green line represents the surgical incision. Pink area corresponds to projection of the scapula on overlying skin. Brown area shows the location of latissimus dorsi muscle.

Simultaneously, the reconstructive team started the elevation of the right STFF. A lazy S incision was made along the mid-axillary line ending in the inferior angle of the scapula (dotted green line, (Figure 2C)). Superior and inferior skin flaps were elevated above the muscular plane. Thereafter, the superior border and the inner aspect of the latissimus dorsi muscle were dissected to expose the scapula tip and identify the angular branch (Figure 3A). To facilitate the isolation of the flap and its pedicle in cephalad direction, the teres major and rhomboid muscles were divided with Harmonic scalpel. The subperiosteal branch of the

circumflex scapular artery was also identified and dissected in cephalad direction (Figure 3A). These two scapular pedicles were fully freed from surrounding tissues in retrograde direction until they converged into a main pedicle (12 cm in length) formed by the subscapular artery and a large accompanying vein. Subsequently, using prefabricated cutting guides and oscillating saw, we performed the bony cuts to obtain the vascularized STFF (Figure 3B). A nonvascularized bony segment was also obtained from the lateral border of the scapula (Figure 2A). Subsequently, under the microscope, end-to-end arterial anastomosis with right facial artery (2.5 mm in diameter) and end-to-side venous anastomosis with right external jugular vein were performed using interrupted stitches of 8-0 nylon and 3.5 mm coupler, respectively.

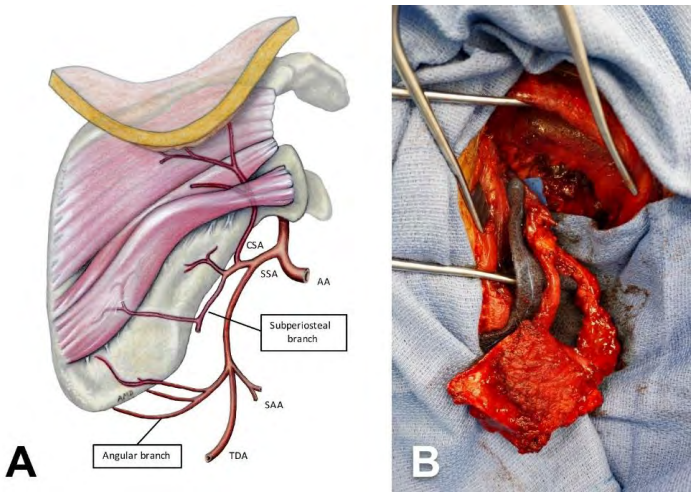

Figure 3(A,B): (A) Schematic representation of vascular anatomy for Scapular Tip Free Flap (STFF) based on angular branch of the Thoracodorsal Artery (TDA) and/or subperiosteal branch of the Circumflex Scapular Artery (CSA). Axillary artery (AA); Subscapular Artery (SSA); serratus anterior artery (SAA). **(B)** Intraoperative image of right scapular tip free flap based on subscapular vessels (pedicle of 12-cm in length). Both angular and periosteal branches provided blood supply to the flap. Length of stay of 6 day. No postoperative complications.

After completion the microvascular step, the flap was inset using the prefabricated plate and monocortical screws (Figure 4A). In addition, as planned, the nonvascularized bone graft was utilized to reconstruct the remaining maxillary-zygomatic defect. This was fixed in position with a separate straight plate and monocortical screws. Next, the oral cavity and neck were profusely irrigated with normal saline and closed in standard fashion. At the right scapular region, the surgical field was also irrigated and hemostasis achieved with bipolar electrocautery. To prevent winged scapula, several holes were drilled through the scapula near to the new post-osteotomy lateral edge. These bony holes were used to reattach previously divided teres major muscle with several stitches of #0 PDS. Two 19 Fr Blake drains were placed inside the surgical cavity, exited through the skin and secured with 3-0 nylon stitches. The incision was finally closed by layers using multiple stitches of 2-0 and 3-0 PDS. The skin closure was accomplished with interrupted stitches of 3-0 nylon. At the end of the case, the patient was transferred to surgical intensive care unit in stable condition. The patient stayed intubated, sedated and in respiratory support until the following morning. The immediate postoperative period was satisfactory without any complications. The histopathologic examination of the specimen reported unicystic (2.9 x 1.4 x 1.1 cm) ameloblastoma with mural invasion. All margins were negative for tumor. Patient was discharged on day 6 after surgery with arm sling to limit Range of Motion (ROM) of the right shoulder. The active ROM of the shoulder was initiated on postoperative day 11 and weight bearing exercises 7 weeks after surgery. Three weeks after surgery, the flap required bulking procedure in clinic due to redundant intraoral soft tissue that was interfering with occlusion (Figure 4(B-C)).

Fourteen months after surgery, patient has no signs of local or systemic recurrence. He reached a satisfactory outcome, working without restrictions with no local pain or edema, inconspicuous scar on the neck, symmetric face, normal MOI and adequate bony structure and intraoral lining for dental implants (Figure 4(C-F)). The donor site of the flap exhibited soft, pliable and painless mature scar. The right upper extremity displayed normal ROM and strength without pain (Figure 5). Patient verbalized to experience only sporadic tingling on the right arm that does not affect his activities of daily living (DASH score of 0.8/100).

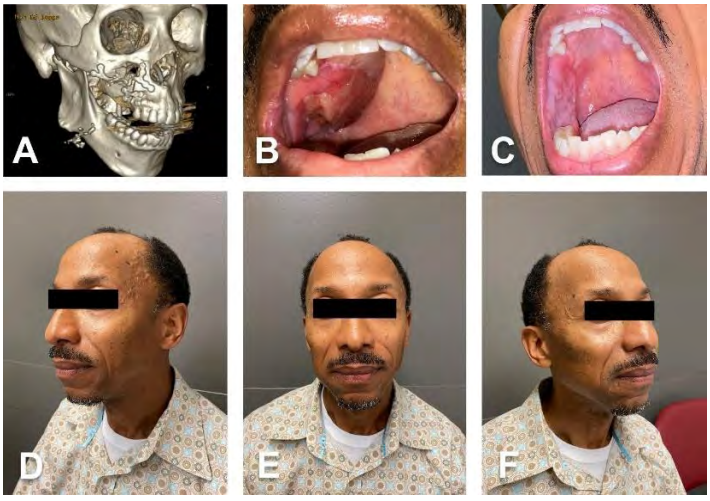

Figure 4 (A-F): (A) Postoperative 3-D reconstruction of right posterior maxillary ameloblastoma treated with scapular tip free flap plus non-vascularized bone graft. **(B)** Early intraoral result showing redundant soft tissue from the STFF interfering with occlusion. **(C)** Long-term intraoral result after flap debulking, normal Maximum Interincisal Opening (MIO) and intraoral lining ready for dental implants' placement. **(D-F)** Long-term cosmetic outcome with symmetric facial appearance.

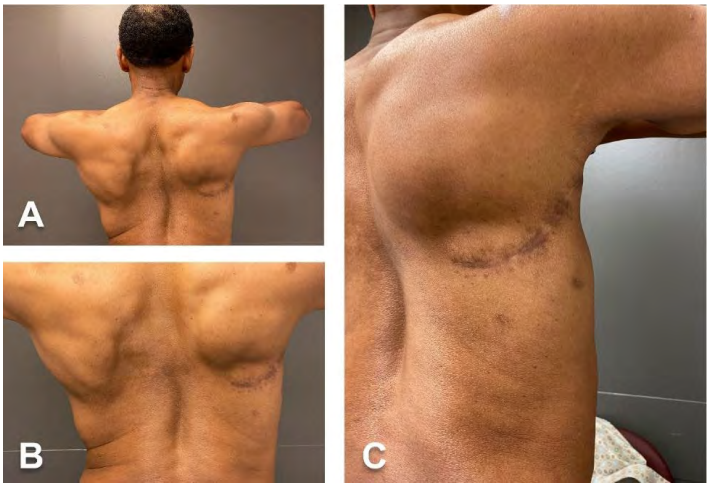

Figure 5: (A-C) Long-term outcome of the donor site of scapular tip free flap. Patient recovered full range of motion and strength of right upper extremity. Only sporadic tingling on arm that does not interfere with his activities of daily living (DASH score of 0.8/100). He developed soft and pliable mature scar. Of note, patient presents congenital scoliosis.

Discussion

Currently, the reconstruction of head and neck tumors needs to consider not only the repair of ablative defects, but also meet expectations of optimal functional and cosmetic outcomes with short hospital stay and minimal complications and residual morbidities. In general, the curative management of ameloblastomas should include the complete excision of the tumor with a 1-1.5-cm safety margin of uninvolved bone around the lesion. This radical approach is often necessary because the definitive diagnosis (including microscopic extension) is only possible after histological examination of the entire specimen [3,16].

The incidence of maxillary ameloblastomas is lower (15%) than their mandibular counterparts [13,14]. However, due to the cancellous nature of the bony structure at their more frequent location (molar region), maxillary ameloblastomas tend to break the "Eggshell-Like" thin cortical bone and subsequently invade adjacent anatomical spaces. For instance, they can spread into pterygomaxillary space, nasal cavity, paranasal sinuses, infratemporal fossa and orbital cavity. Furthermore, maxillary ameloblastomas can infiltrate the skull base, and even the central nervous system which account for most of fatal cases of this condition [12-14]. Accordingly, radical resection of any maxillary ameloblastoma (i.e., partial/total maxillectomy, *en bloc* resection) seems to be necessary to ensure local control of the disease. However, the deep anatomical confinement of these lesions and proximity of vital structures may cause highly challenging resections and often insufficient safety margins [15]. For this reason, maxillary ameloblastomas present elevated postoperative complications as well as significantly higher local and distant recurrence rate [15]. Indeed, radical tumor resection in these areas generate secondary defects that require complex reconstructive procedures. Thus, immediate reconstruction with vascularized bone grafts have acquired popularity among surgeons due to psychosocial benefits of one-stage surgeries and the advantage of performing microsurgical procedures in absence of tissue fibrosis or scar contracture. This approach not only restore the bone structure, but also favors early intervention on functional aspects of the surgical reconstruction such as speech, deglutition, mastication and dental rehabilitation.

In this regard, even though it is not usually the first choice for maxillary reconstruction, Scapular Tip Free Flaps (STFF) have multiple advantages such as reliable vascular anatomy, long pedicle, several chimeric options and low rate of donor site morbidity. Their versatility makes these flaps suitable for a large variety of surgical settings. Furthermore, the muscular component (cuff of teres major muscle) can be mucosalized in few weeks and provide adherent mucosa lining on the alveolar ridge for dental implants placement. Accordingly, our patient got an uneventful surgery and short hospital stay with a satisfactory long-term cosmetic and functional outcome without major complications. He recovered full ROM and strength of the right upper extremity and returned to work in the same position as prior to the surgery and without restrictions. He is currently in the process of oral rehabilitation pursuing the placement of dental implants. After 14 months post-surgery, our patient is free of local or systemic recurrence [1,7]. Because of relapses mainly occur within the first 5 years after the primary surgery, we are planning to continue regular follow-up with this patient for at least 4 more years.

Conclusions

The anatomical confinement and easy spread to surrounding tissues make difficult the successful resection of maxillary ameloblastomas and increase the risks of postoperative complications and recurrence. Therefore, an experienced multidisciplinary team is required to ensure appropriate outcomes not only from ablative and reconstructive points of view, but also from the functional and psychological aspects of these cases. Reconstructive procedures for maxillary ameloblastomas with vascularized bone grafts such as fibula free flap and STFF have demonstrated excellent cosmetic and functional results.

Disclosure Statement

The authors declare that there is no conflict of interest.

References

1. Olaitan AA, Arole G, Adekeye EO (1998) Recurrent ameloblastoma of the jaws. Int J Oral Maxillofac Surg 27: 456-460.
2. Rapidis AD, Andressakis DD, Stavrianos SD, Faratzis G, Arnogiannaki-Liappi N, Lagogiannis GA, et al. (2004). Ameloblastomas of the jaws: clinico-pathological review of 11 patients. Eur J Surg Oncol 30: 998-1002.
3. Ghandhi D, Ayoub AF, Pogrel MA, MacDonald G, Broklebank LM, et al. (2006) Ameloblastoma: a surgeon's dilemma. J Oral Maxillofac Surg 64: 1010-1014.
4. Fregnani ER, da Cruz Perez DE, de Almeida OP, Kowalski LP, Soares FA, et al. (2010) Clinicopathological study and treatment outcomes of 121 cases of ameloblastomas. Int J Oral Maxillofac Surg 39: 145-9.
5. Hertog D, Bloemena E, Aartman IHA, van-der-Waal I (2012) Histopathology of ameloblastoma of the jaws; some critical observations based on a 40 years single institution experience. Med Oral Patol Oral Cir Bucal 17: e76-e82.
6. Hong J, Yun PY, Chung IH, Myoung H, Suh JD, et al. (2007) Long-term follow up on recurrence of 305 ameloblastoma cases. Int J Oral Maxillofac Surg 36: 283-288.
7. Reichart PA, Philipsen HP, Sonner S (1995) Ameloblastoma: biological profile of 3677 cases. Eur J Cancer B Oral Oncol 31B: 86-99.
8. Hosalkar R, Saluja TS, Swain N, Singh SK (2020) Prognostic evaluation of metastasizing ameloblastoma: A systematic review of reported cases in literature. J Stomatol Oral Maxillofac Surg S2468-7855(20)30164-6.
9. Gunaratne DA, Coleman HG, Lim L, Morgan GJ (2015) Ameloblastic carcinoma. Am J Case Rep 16: 415-419.
10. Gardner DG (1984) A pathologist's approach to the treatment of ameloblastoma. J Oral Maxillofac Surg 42: 161-166.
11. Yang R, Liu Z, Peng C, Cao W, Ji T (2017) Maxillary ameloblastoma: Factors associated with risk of recurrence. Head Neck 39: 996-1000.
12. Nastri AL, Wiesenfeld D, Radden BG, Eveson J, Scully C (1995) Maxillary ameloblastoma: a retrospective study of 13 cases. Br J Oral Maxillofac Surg 33: 28-32.
13. Evangelou Z, Zarachi A, Dumollard JM, Peoc'h M, Komnos I, et al. (2020) Maxillary ameloblastoma: A review with clinical, histological and prognostic data of a rare yumor. *In Vivo* 34: 2249-2258.
14. Parmar S, Al-Qamachi L, Aga H (2016) Ameloblastomas of the mandible and maxilla. Curr Opin Otolaryngol Head Neck Surg. 2016; 24: 148-154.
15. Petrovic ID, Migliacci J, Ganly I, Patel S, Xu B, et al. (2018) Ameloblastomas of the mandible and maxilla. Ear Nose Throat J 97: E26-E32.
16. Pogrel MA, Montes DM (2009) Is there a role for enucleation in the management of ameloblastoma? Int J Oral Maxillofac Surg 38: 807-812.