Resection of a Large, Central Hemangioma With Reconstruction Using a Radial Forearm Flap Combined With Zygomatic and Pterygoid Implants

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Cavernous hemangiomas are rare, benign proliferations of vessels that usually occur at or just after birth. Few cases have been reported in the literature, however, involving the maxilla. We report a rare case of central cavernous hemangioma of the maxilla in an elderly male, review the literature, and discuss treatment options. An algorithm for treatment of all vascular lesions has been developed based on current literature, modern imaging and endovascular technology. With proper diagnosis, planning and treatment, large vascular lesions can be managed with minimal blood loss and morbidity. This patient was reconstructed to full function utilizing zygomaticus implants and an implant-retained prosthesis.

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Cavernous hemangiomas are rare, benign proliferations of vessels that usually occur at or just after birth. They are usually masses in the parotid or in the skin over the parotid region. When they occur centrally, they are most commonly found in the posterior mandible,1 although several cases have been reported within the zygoma. There is some controversy as to whether this entity is a hamartoma or true tumor.1 Few cases have been reported in the literature, however, involving the maxilla, especially in an elderly man. We report a rare case of a central cavernous hemangioma in an elderly man, review the literature, and discuss treatment options.

Report of a Case

A 77-year-old man presented to a local oral and maxillofacial surgeon with a complaint of recent onset of sinus congestion and frequent epistaxis, 1 episode of which required hospitalization and transfusion. The patient also reported a history of a slow-growing painless mass in the maxilla that had been present for 1 year.

Physical examination showed a left palatal mass measuring approximately 6 × 4 cm (Fig 1). The mass was firm and nontender, with intact mucosa. There were no palpable thrills. Buccal expansion was also noted from the right central incisor to the left molar (Fig 2). The dentition was nonmobile and nontender to percussion. Needle aspiration of the lesion initially showed bright red blood followed by dark blood. A panoramic radiograph showed a large radio-opaque lesion of the left maxilla from the midline to the maxillary buttress.

FIGURE 1. Palatal mass.

A computed tomography scan with contrast was obtained and showed a left maxillary expansile lesion. The lesion was found to have a soap-bubble appearance (Fig 3).

An incisional biopsy specimen was obtained in the operating room (at an outside institution) for control of potential bleeding because a vascular lesion was suspected. Results of the biopsy were consistent with a vascular lesion with the differential including a cavernous hemangioma versus an arteriovenous malformation. The patient was subsequently referred to a university setting for further treatment. A magnetic resonance angiography scan of the face and neck was obtained and showed a large maxillary lesion with small feeder vessels but no large vascular inflow and outflow. A preliminary diagnosis of central cavernous hemangioma was made.

A discussion was held with the patient regarding treatment and reconstruction options. The treatment plan included an angiogram with selective embolization of the appropriate feeding vessels, followed by subtotal maxillectomy with a radial forearm free flap for closure of the defect. The evening preceding the surgical procedure, the patient underwent angiography. The lesion was confirmed to be a low-flow vascular lesion (Fig 4). During the procedure, embolization of the lesion was performed by placing a coil in the left internal maxillary artery as well further distally in branches feeding the palate and nose (Fig 5).

The next morning, the patient was taken to the operating room for definitive treatment. A Weber-Ferguson incision was performed for access to the maxillary le-
sion. A partial maxillectomy was performed from the right central incisor to the left second molar region. The specimen was fully mobilized and removed with no untoward bleeding (Figs 6, 7). The specimen was sent to the pathology department, where a definitive diagnosis of cavernous hemangioma was obtained through frozen section. A radial forearm fasciocutaneous free flap was then harvested with a 6 × 4–cm portion for coverage of the maxillary defect and vascular pedicle of approximately 12 cm. The pedicle was carefully tunneled from the maxilla through the parapharyngeal space to the neck, where anastomosis was completed by use of the facial artery and vein. The flap was then inset to the maxilla. In addition, a portion of the flap was de-epithelialized and folded into the nasal cavity for internal resurfacing. The patient’s postoperative course was uneventful. He recovered well and without complication. The patient underwent subsequent restoration with standard endosteal, pterygoid, and zygomatic implants at 4 months (Figs 8-10) and had a final prosthesis placed with bilateral bars for support at 10 months postoperatively (Figs 11, 12).

Discussion

In 1982 Mulliken and Glowacki described a clinically relevant classification of vascular anomalies in which these lesions were categorized according to
their endothelial characteristics. This classification, which was adopted by the International Society for the Study of Vascular Anomalies in 1996, differentiates proliferating tumors (most of which are hemangiomas) from vascular malformations, which are structural anomalies involving capillaries, venules, veins, lymphatic channels, and combinations of these structures (Table 1). Hemangiomas are characterized histologically by high endothelial cell turnover, and their clinical “life cycle” includes proliferative, plateau, and involution phases. Hemangiomas are further characterized by cell markers (GLUT-1, merosin, Lewis Y) that are otherwise found only in human placental tissue. Most other vascular anomalies are properly termed malformations, which are characterized by normal endothelial cell turnover and abnormal gross vascular anatomy. These include high-flow lesions (which have an arterial component) and low-flow lesions (capillary and venous).2

Hemangiomas are benign lesions that rarely affect bone, in contrast to vascular malformations, which affect bone in 35% of all cases.3 Central hemangiomas compose less than 1% of all bone lesions, and when they do occur, they almost always appear in the facial bones and skull.4 They occur most frequently in the

![Figure 8](image8.png)

**FIGURE 8.** A, B, Four months postoperatively.


![Figure 9](image9.png)

**FIGURE 9.** Implant insertion: endosteal implants in right maxilla, 1 pterygoid implant in left posterior region, and 1 zygomaticus implant in left zygoma.


![Figure 10](image10.png)

**FIGURE 10.** Postoperative panoramic radiograph.

Hemangiomas may be radiosensitive, although few authors have reported success. In 1 case, the decision was made to treat a central hemangioma of the maxilla with 3,000 rad of radiation therapy because during a biopsy, transfusion was required as a result of severe hemorrhage. The patient was reported to be doing well during follow-up; however, good long-term follow-up with objective outcome measurements has not been provided. In 1 case report, a 36-year-old man with a right maxillary sinus hemangioma could not be treated with embolization because the lesion relied on the right ophthalmic artery, and surgical treatment was not amenable because there was significant bleeding encountered during an initial surgical attempt. Therefore the authors decided on radiation therapy consisting of a total dose of 50 Gy. Magnetic resonance imaging at 1 month showed a small reduction. The lesion continued to decrease in size up to 6 months, at which time there was a 50% reduction in size and the patient was clinically asymptomatic. At final follow-up at 2 years, it was reported that the tumor reduction had been maintained without reappearance of the initial symptoms. There appear to be palliative effects of radiation without complete eradication of the lesion. The risks of radiation therapy are not without merit, including radiation telangiectasis and radiation-induced neoplasms. One case report combined radiation with surgery. In this case a 19-year-old woman with a hemangioma of the right maxilla was treated with 5,000 rad over a period of 6 months, with some reduction in tumor size. This was followed by ligation of the external carotid artery and surgical removal of residual tumor accessed via a Weber-Ferguson incision.

A second treatment option has been injection of sclerosing agents, such as sodium morrhuate, sodium psyllate, or 250 mg of tetracycline in 5 mL of saline.

FIGURE 11. Bilateral bar placement for prosthetic support.

FIGURE 12. Final prosthetic result.
solution. Outcomes are considered unpredictable and range from minimal effect on the hemangioma to over-fibrosis. A recent study in the literature has used pingamycin injection for cavernous hemangiomas, reporting a curative rate of approximately 94%. These lesions, however, all have been in soft tissue; hence the effectiveness on intraosseous lesions needs to be further studied.

Surgical treatment of central cavernous hemangiomas requires 2- to 3-mm peripheral margins. Smaller lesions have been reported to be surgically excised with curettage. Larger lesions have required wider excision, although ablation with cryosurgery or laser surgery reportedly is possible. For surgical access to the maxilla, smaller lesions have been accessed via a Caldwell-Luc approach, where large lesions have been best accessed via Weber-Ferguson incisions. Another treatment option has been embolization, with or without surgery. It is not an entirely benign procedure, with risks including reflux of emboli into the internal carotid artery as well as possible adverse ischemia of supplied areas. In 1 report, embolization was not carried out because the lesion was supplied by the ophthalmic artery. Embolization alone has been used in cases in which arteriovenous malformations have been diagnosed, whereas cavernous heman-

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**FIGURE 13.** Treatment algorithm for vascular lesions. CT, computed tomography; MRA, magnetic resonance angiography; Tx, treatment; Angio, angiography; dx, diagnosis.

Angiomas have mostly been treated by a combination of embolization and surgery. In these cases, a group of authors suggests that because of the excellent blood supply in the facial region, surgery must be done within 36 hours to avoid collateralization. There was a second report of an elderly man with a cavernous hemangioma of the maxilla who underwent embolization of the feeding vessels, followed by ligation of the external carotid artery before surgical excision of the lesion accessed via the Weber-Ferguson incision. There was lack of hemorrhage, although this cannot provide definitive proof that the embolization helped because the authors also ligated the external carotid artery.

When one is treating central hemangiomas, good preoperative planning is critical in avoiding disastrous surgical consequences. The surgeon should have a thorough knowledge of all vascular lesions and the step-by-step workup of these lesions. An algorithm that we use is presented in Figure 13. This includes obtaining a magnetic resonance angiography scan upon suspicion of a vascular lesion, allowing for differentiation between high-flow and low-flow lesions. Low-flow lesions can undergo surgical treatment alone. The addition of embolization can be used for very large low-flow lesions to decrease blood loss. High-flow lesions can undergo embolization alone followed by surgery if they are nonresolving. Mixed high-flow/low-flow lesions should undergo embolization followed by surgical treatment because of the low probability of involution after embolization.

Perioperative preparation should include the availability of blood as well as the ability to ligate feeding blood vessels. The most impressive reduction of the risk of hemorrhage seems to result from preoperative embolization. However, as reported by Perugini et al., one must perform surgery fairly soon after the angioscopy and embolization to avoid recollateralization.

Several options are available for palatomaxillary re-construction; however, this is beyond the scope of our discussion. We chose a combination of a microvascular free flap for an oral/nasal seal in combination with zygomatic and pterygoid implants in lieu of bony reconstruction.

References