

# Arteriovenous Fistula of the Lower Lip: Case Report of Combined Intra-Vascular and Surgical Cure

Timothy J. Martin, MD; Lotfi Haccin-Bey, MD; John S. Rhee, MD, MPH

## ABSTRACT

*Purpose:* We describe a patient presenting with a vascular mass of the lower lip with a history of traumatic lip-biting. The lesion was treated with preoperative intravascular embolic therapy and surgical excision.

*Summary:* Arteriovenous fistula (AVF) of the head and neck are vascular lesions with a single connection between the involved artery and vein. Trauma to the area, often in the distant past, is often seen as the inciting event. We describe a patient with a lower-lip AVF with repeated episodes of lip biting that caused expansion of the mass. The patient underwent preoperative embolic therapy and surgical excision with excellent functional and cosmetic outcome.

*Conclusion:* Arteriovenous fistula of the lower lip can be successfully managed with preservation of lip function and cosmesis through combined intravascular and surgical therapy.

## INTRODUCTION

Malformations of vascular structures present both diagnostic and therapeutic challenges. When the malformation occurs in the head and neck region, particularly the face, the challenge expands to include the preservation of both functional concerns and cosmetic appearance of the affected structures.

Vascular malformations have been known as a clinical entity since the mid-18th century. Traumatic arteriovenous malformations were recognized as more frequent occurrences during major armed conflicts of

the 20th century.<sup>1</sup> The pathophysiology of AVF and why they occur with traumatic injury is not well-understood. With the advent of modern arteriography, precise localization of the source arterial vessel became possible, making excision and reconstructive planning more precise.<sup>1-2</sup> The management of these abnormalities continues to expand with the use of computed tomography (CT) angiography and intravascular embolic therapies. As the techniques for removal have advanced, the need for reconstructive techniques has evolved accordingly.

Arteriovenous fistula (AVF) is defined as a vascular lesion having a single connection between an artery and a vein.<sup>3</sup> It is distinct from arteriovenous malformations (AVM), which are congenital and often large, disfiguring lesions with numerous regions of connectivity. The majority of AVMs reported in the literature are congenital in origin and lack the traumatic origin that is identifiable in the natural history of AVF.<sup>4</sup> We report an AVF of the lower lip that was successfully managed with state-of-the-art combined embolic and surgical therapy.

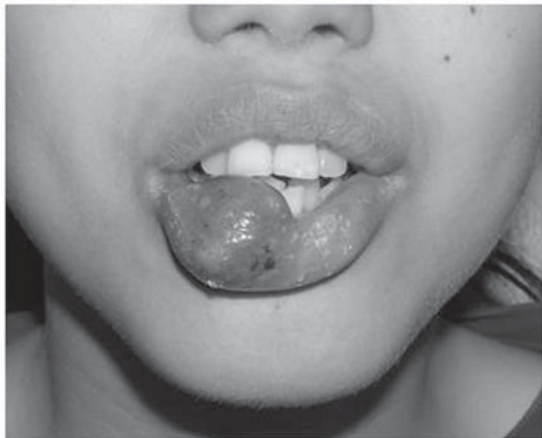
## CASE REPORT

A 19-year-old female presented with a 7-year history of a slowly growing right lower lip lesion. The lesion began following traumatic lip biting and had subsequently grown on a gradual basis with occasional bleeding after accidental injury. She denied any significant pain, but was having trouble sealing her lips to eat and drink.

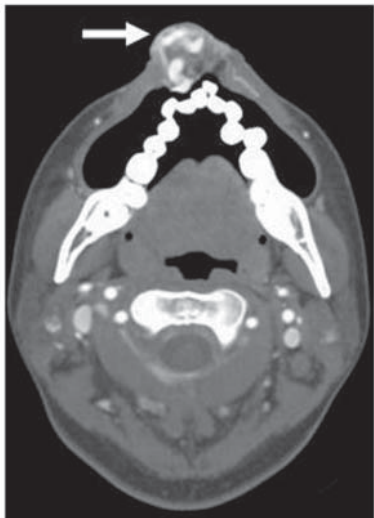
On examination, the mass was pulsatile, encompassing the right side of the lower lip extending close to the midline, but sparing the oral commissure. The lesion extended onto the cutaneous portion of the lower lip, as well as onto the intra-oral aspect (Figure 1.)

In light of the pulsatile nature of the lesion, CT angiographic evaluation was done to identify the nature of the lesion and source vessels. The CT angiogram

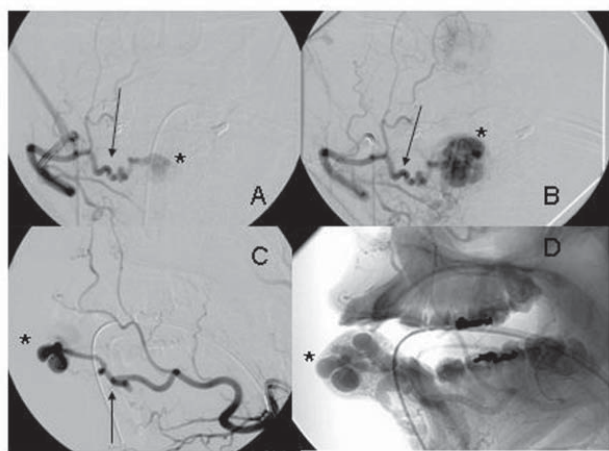
Authors are with the Medical College of Wisconsin, Milwaukee, Wis. Doctors Martin and Rhee are with the Departments of Otolaryngology and Communication Sciences and Doctor Haccin-Bey is with the Department of Radiology. Please address correspondence to: Timothy J. Martin, MD, Department of Otolaryngology and Communication Sciences, Medical College of Wisconsin, 9200 W Wisconsin Ave, Milwaukee, WI 53226; phone 414.895.5580; fax 414.805.7890; e-mail tmartin@mcw.edu.



**Figure 1.** Lower lip mass seen at presentation in a 19-year-old female.



**Figure 2.** CT angiogram demonstrating an AVF 2 cm in diameter (arrow).



**Figure 3.** A and B Antero-posterior arteriogram demonstrating an AVF (\*) supplied by the inferior labial artery prior to embolization (arrow). C and D Lateral arteriogram demonstrating an AVF (\*) with the inferior labial artery highlighted by an arrow.

revealed an AVF, measuring 2 cm in diameter (Figure 2). The AVF was supplied mainly by a dilated right facial artery with multiple tortuous draining veins. Given the size and relationship to the facial artery, pre-surgical embolization was performed by a neuro-interventional radiologist. Surgery was coordinated to follow embolization.

During the embolization procedure, the ectatic and tortuous right inferior labial artery was superselectively catheterized and embolized with liquid polymer (Figure 3). The left inferior labial artery showed no abnormality. Post-embolic angiography demonstrated near-complete obliteration of the right inferior labial artery (Figure 4). In the post-embolic period, prior to surgical therapy, the lesion was reduced to approximately half its pre-embolic size. No bleeding or embolic complications occurred.

Given the bulk of the remaining mass, the likelihood of accidental biting reinjury, and the patient's difficulty with oral competence, resection of the lesion was performed under monitored anesthesia 2 days after embolization. An incision was made overlying the mass along the red portion of the lip. The necrotic overlying mucosa was excised, along with the lesion in an elliptical fashion. The remnant AVF was excised from the surrounding tissue. The mass measured approximately 3.0 x 2.5 cm. The thrombosed vein and artery were identified. The mass was taken en bloc and sent for pathological review. The margins of the pathologic specimen were reported as microscopically clear of vascular abnormality.

In order to close the defect in an ideal fashion, a minimal wedge-type of excision was made that created a through-and-through defect. Part of the orbicularis oris muscle was excised as well, as it was noted to be quite attenuated by the tumor. Next, the orbicularis oris muscle was reapproximated and the vermilion border was realigned. The remaining mucosa and skin were closed in layers (Figure 5).

In the post-operative period, the surgical site healed well despite a small wound infection that required a brief course of antibiotics. The vermilion was well aligned. The lip contour appeared excellent, especially anterior to the wet line. A small amount of residual fullness behind the wet line was present at 6 weeks. The muscle function appeared to be symmetric and normal (Figure 6). The cosmetic appearance was satisfactory to both the patient and the surgeon.

**COMMENT**

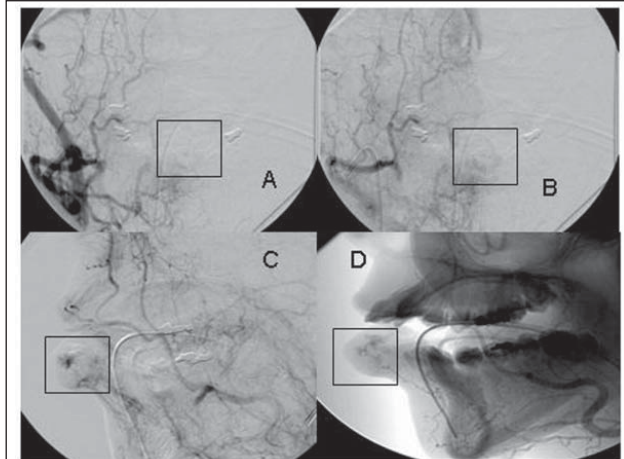
Acquired arteriovenous fistula has been defined as a le-

sion having a single communication between an artery and a vein.<sup>3</sup> It is a distinct clinical entity from congenital AVM, which has multiple distinct connections and is often massive in size. AVFs of traumatic origin have been reported in all regions of the face. The lesions are often associated with high velocity blunt traumatic injury, including automobile collision, falls from elevation, and frequently as a result of gloved-fist boxing.<sup>1-3,6,7</sup> One previous report of a lower lip AVF was found, with the inciting trauma from a baseball.<sup>7</sup> No previous report of lip biting as the inciting event for an AVF could be found.

The standard diagnostic approach to an AVF begins with clinical suspicion from exam findings. A palpable thrill or auscultated bruit over the lesion belies its vascular nature. Compressing the proximal arterial supply of a high-flow AVF may actually result in reduction of heart rate, a phenomenon known as Branham-Nicoladoni sign.<sup>6</sup>

Proceeding to CT angiographic evaluation from that point not only serves to confirm the clinical diagnosis but begins therapeutic planning. CT angiography will identify feeding vessels and determine the possibility of embolic therapy. In our case, CT angiography elicited the arterial nature of the lesion and determined that a dilated facial artery was the feeding vessel. The dilated venous drainage pathway resulted from the increased vascular pressure transmitted to the highly compliant venous system.

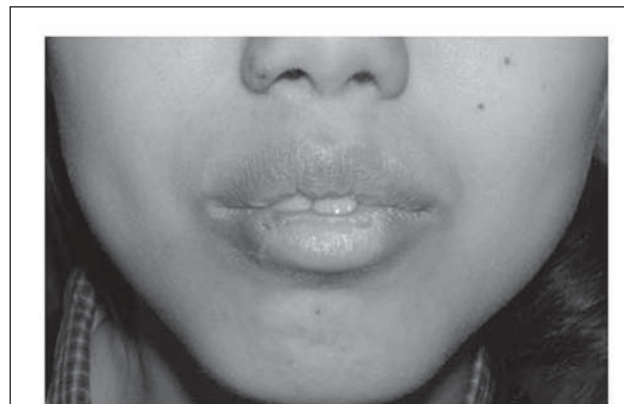
Combined embolic and surgical therapy was undertaken in this case for several reasons. Embolization of the single connection between the inferior labial artery and vein was extremely effective in stopping the dominant blood flow to the lesion. If embolic therapy alone had been performed, the patient would have been left with a firm mass of coagulum in the substance of the lower lip. In other less conspicuous parts of the body, this may have been an acceptable outcome. However, the dynamic and highly visible nature of the lower lip makes any small contour abnormality easily visible with concomitant adverse functional consequences. Additionally, since the inciting event was lip biting, the presence of a mass beneath the lip surface would increase the likelihood of reinjury. Surgical resection without preoperative embolization would likely have had a greater amount of blood loss and required the sacrifice of a greater amount of normal tissue. The loss of more normal tissue in the face would have likely produced a less pleasing cosmetic outcome and could have resulted in symptomatic microstomia.



**Figure 4.** A and B antero-posterior post-embolic arteriogram shows near complete obliteration of the AVF highlighted by the absence of flow in the boxed area. C and D lateral post-embolic arteriogram.



**Figure 5.** Intra-operative photo after AVF excision and reconstruction.



**Figure 6.** Three-month post-operative photo after combined intravascular and surgical treatment.

## CONCLUSION

Current state-of-the-art management for a case of lower lip arteriovenous fistula is presented. Combined embolization and surgical resection resulted in complete excision with an optimal cosmetic and functional outcome.

## REFERENCES

1. Schwartz GF, Rankow RM. Traumatic arteriovenous fistula of the facial artery. *Plas Reconstr Surg.* 1967;40(5):453-456.
2. Holt GR, Holt JE, Cortez EA, Thornton WR, Young WC. Traumatic facial arteriovenous malformations. *Laryngoscope.* 1980;90(12):2011-2020.
3. van Sant TE Jr, Creely JJ Jr. Traumatic arteriovenous fistula of the external nose. *Arch Otolaryngology.* 1974;99(2):145-146.
4. Kohout MP, Hansen M, Pribaz JJ, Mullikan JB. Arteriovenous malformations of the head and neck: natural history and management. *Plas Reconstr Surg.* 1998;102(3):643-654.
5. Stucker FJ. Extracranial arteriovenous fistulas. *Laryngoscope.* 1974;84:970-975.
6. Rance BR, Laws RA, Keeling JH 3rd, Warden PJ. Traumatic arteriovenous fistula of the upper lip. *Cutis.* 1998;62(5):235-237.
7. Marks MW, Argenta LC, Dingman RO. Traumatic arteriovenous malformation of the external carotid arterial system. *Head Neck Surg.* 1984;6(6):1054-1058.

# Wisconsin Medical Journal

The mission of the *Wisconsin Medical Journal* is to provide a vehicle for professional communication and continuing education of Wisconsin physicians.

The *Wisconsin Medical Journal* (ISSN 1098-1861) is the official publication of the Wisconsin Medical Society and is devoted to the interests of the medical profession and health care in Wisconsin. The managing editor is responsible for overseeing the production, business operation and contents of the *Wisconsin Medical Journal*. The editorial board, chaired by the medical editor, solicits and peer reviews all scientific articles; it does not screen public health, socioeconomic or organizational articles. Although letters to the editor are reviewed by the medical editor, all signed expressions of opinion belong to the author(s) for which neither the *Wisconsin Medical Journal* nor the Society take responsibility. The *Wisconsin Medical Journal* is indexed in Index Medicus, Hospital Literature Index and Cambridge Scientific Abstracts.

For reprints of this article, contact the *Wisconsin Medical Journal* at 866.442.3800 or e-mail [wmj@wismed.org](mailto:wmj@wismed.org).

© 2006 Wisconsin Medical Society