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CASE REPORT
Craniofacial/Pediatric

A Simple Surgical Solution for Functional Improvement of Deforming Vascular Malformations with Lip Involvement

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Summary: The management of large, deforming facial arteriovenous malformations (AVMs) can be a daunting clinical challenge for patients and surgeons. Many patients delay treatment due to fear of surgical intervention and concern for unfavorable cosmetic outcomes. Delaying treatment can lead to soft-tissue hypertrophy. Occasionally, facial AVMs can also present with lip involvement, necessitating surgical intervention. A major potential issue regarding AVMs managed with surgical excision is excessive bleeding due to the enlarged dermal vascular plexus. Here, we present a simple surgical technique used to manage deforming AVMs involving the lips. A 32-year-old man with Sturge-Weber syndrome and a 72-year-old man with AVMs of the left face presented to clinic for management. The malformations involved the lips in both patients. Both patients had constant drooling and difficulty eating and talking, in addition to aesthetic concerns. They both underwent surgical excision of the redundant tissue with minimal undermining, advancement flaps, and layered closure. Minimal undermining allowed successful removal of the AVMs and redundant tissue without excessive blood loss and allowed healing without complications. Both patients had significant improvement in eating and talking without drooling. Aesthetically, satisfactory outcomes were maintained at 1-year follow-up appointments. Removal of AVMs while minimizing blood loss can be a challenging endeavor. Here, we have presented a successful, effective technique for restoring both form and function in patients with AVMs involving the lips, with good long-term results. *(Plast Reconstr Surg Glob Open 2023; 11:e4816; doi: 10.1097/GOX.0000000000004816; Published online 17 February 2023.)*

Vascular anomalies of the face can broadly be categorized into two subtypes: vascular neoplasms and vascular malformations.[1] Vascular neoplasms tend to demonstrate a pattern of growth followed by regression. Vascular malformations are typically present at birth and grow at a steady rate over time.[2] Many treatments for facial vascular malformations have been proposed, including oral steroids, sclerotherapy, cryotherapy, interferon therapy, cosmetic tattooing, photodynamic therapy, laser treatment, embolization, and surgery.[3–6] Surgical treatment is typically reserved for lesions that interfere with eating, involve the lips, are larger than 2 cm^3, or have associated soft-tissue hypertrophy.[3,4,7]

Management of large, deforming arteriovenous malformations (AVM) can be a clinical challenge for surgeons and a major burden for patients. Depending on the location and size of these malformations, patients can experience significant functional, aesthetic, social, and psychological consequences. When the lips are involved, additional deficits in speech, feeding, and excessive drooling may be present.[4] Facial AVMs also create aesthetic issues, including facial asymmetry with animation, imbalance of cosmetic facial subunits, disruption of hair follicles, skin discoloration, and poor skin quality.

Large vascular malformations should ideally be treated early to reduce functional and aesthetic complications. Unfortunately, many patients delay treatment due to fear

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of intervention or difficulties with access to medical or surgical care. When treatment is delayed, the lesions continue to grow with concomitant soft-tissue hypertrophy and potential for bony remodeling, increasing the complexity of potential surgical management. Here, we present the surgical approach successfully used in two patients with delayed presentation of AVMs with lip involvement.

CASE PRESENTATION

The first patient was a 32-year-old-man with Sturge-Weber syndrome who presented with vascular malformations involving most of his face, including his upper and lower lips (Fig. 1). The patient underwent debulking surgery at an outside hospital several years before presentation. Despite this management, the AVMs continued to grow, rapidly reaching disfiguring dimensions. These disfiguring malformations created significant difficulty with eating and caused excessive drooling. On examination, the lesions encompassed 50% of the upper lip and 100% of the lower lip.

The second patient was a 72-year-old man with a congenital vascular malformation affecting the left side of his face and neck, with disfiguring enlargement of the lower lip and left ear (Fig. 2). The patient received no prior treatment before presentation. His AVM created a severe deformity with oral incompetence that caused excessive drooling and difficulty talking and eating. On examination, the lesions encompassed 100% of the lower lip and minimally involved the upper lip.

Preoperative angiography was not performed on either patient because the venous component of the malformation did not have any suspicion of high-flow component on physical examination. Both cases were treated with full thickness excision of the redundant tissues with advancement lip flaps for coverage. As mostly redundant lip tissue was excised, no significant reduction of the oral vestibule was generated by the procedure. Minimal mucosal undermining was performed to avoid excessive bleeding. The first patient required a wedge resection of approximately 40% of the upper lip and 60% of the lower lip based on the AVM's area of maximum volume. The second patient required wedge resection of approximately 80% of the lower lip and excision with reconstruction of the left ear lobule and infra-auricular skin (Fig. 1). [**See figure, Supplemental Digital Content 1,** which shows preoperative markings and advancement flap design of patient 2. Right image: the mucosal markings show a preserved mucosal rim at the level of the lower buccal sulcus (white arrow), **http://links.lww.com/PRSGO/C397**.]

Full thickness excision with minimal undermining of the mucosa allowed successful removal of the AVMs and redundant tissue without excessive blood loss. The advancement flaps provided adequate coverage that healed without complication. Both patients had significant improvement in oral competence and were able to eat and talk without drooling. Aesthetically, they were both pleased with the outcome at 6-weeks, and their results were maintained at 1-year follow-up appointments (Figs. 3 and 4).

Fig. 1. Patient 1. A 32-year-old man with Sturge-Weber syndrome with vascular malformations involving upper and lower lips.

Fig. 2. Patient 2. A 72-year-old man with congenital vascular malformation involving the lower lip, left face, and left ear.

Fig. 3. Patient 1. Postoperative results at 6 weeks postoperatively.

Fig. 4. Patient 2. Postoperative results at 6 weeks postoperatively.

DISCUSSION

Patients with delayed presentation of AVMs can pose significant challenges in management. Surgical treatment is often necessary for those involving the lips, with significant soft tissue hypertrophy, or with functional deficit. Surgery is focused on restoring form and improving function such as speech, eating, and oral competence.

The literature addressing the management of complex, deforming AVMs of the face is not substantial. Surgical techniques have been described for AVMs involving the lips, including vertical and horizontal wedge resection, elliptical excision of the vermillion,[8,9] and a modified technique combining the "bikini lip reduction" with the bull horn technique for upper lip lifting.[7,10] The majority of these techniques, however, were described either in pediatric populations or for vascular malformations involving the upper lip specifically. In this case, we found that simple excision with minimal undermining presented the best opportunity for AVM removal without excessive bleeding and significant symptomatic improvement.

Large facial AVMs that involve the lips can be disfiguring and create significant functional deficit. Initially, the treatment strategy may seem intimidating and difficult. Nonetheless, we have demonstrated two cases of disfiguring AVMs involving the lips where excision of redundant tissue with minimal undermining of the flaps proved to be a safe and effective surgical approach.

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All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

PATIENT CONSENT

Informed consent was obtained from all patients for being included in the study. Additional informed consent was obtained from all patients for whom identifying information is included in this article.

REFERENCES

1. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg.* 1982;69:412–420.
2. McCuaig CC. Update on classification and diagnosis of vascular malformations. *Curr Opin Pediatr.* 2017;29:448–454.
3. Van Doorne L, De Maeseneer M, Stricker C, et al. Diagnosis and treatment of vascular lesions of the lip. *Br J Oral Maxillofac Surg.* 2002;40:497–503.
4. Cerrati EW, O TM, Binetter D, et al. Surgical treatment of head and neck port-wine stains by means of a staged zonal approach. *Plast Reconstr Surg.* 2014;134:1003–1012.
5. De Sousa Almeida LM, Dick TNA, Santos LR, et al. Sclerotherapy of a lower lip vascular malformation: case report. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2020;129:e90–e91.
6. Low DW. Management of adult facial vascular anomalies. *Facial Plast Surg.* 2003;19:113–130.
7. Dessy M, Colletti G, Dionisio A, et al. Surgical correction of hypertrophic upper lip in vascular malformations. *J Craniomaxillofac Surg.* 2018;46:1790–1792.
8. Zide BM, Glat PM, Stile FL, et al. Vascular lip enlargement: Part II. Port-wine macrocheilia—tenets of therapy based on normative values. *Plast Reconstr Surg.* 1997;100:1674–1681.
9. Zide BM, Glat PM, Stile FL, et al. Vascular lip enlargement: Part I. Hemangiomas—tenets of therapy. *Plast Reconstr Surg.* 1997;100:1664–1673.
10. Fanous N, Brousseau VJ, Yoskovitch A. The "bikini lip reduction": a detailed approach to hypertrophic lips. *Can J Plast Surg.* 2007;15:205–210.