Maxillofacial rehabilitation of a large facial defect resulting from an arteriovenous malformation utilizing a two-piece prosthesis

Donna M. Hecker, DDS, MS[a]
School of Dentistry, University of Minnesota, Minneapolis, Minn.

Large facial defects involving the oral cavity can be difficult to restore prosthetically because of a lack of anatomic undercuts, limited means of retention, mobility of soft tissue margins, and the weight of the prosthesis. Use of skin adhesives may be precluded because of the presence of persistent moisture and saliva. The maxillofacial rehabilitation, including the design and fabrication of a 2-piece silicone prosthesis retained by the teeth, of a patient with a large facial defect as a result of treatment for an arteriovenous malformation is described. The pathogenesis and therapeutic alternatives for arteriovenous malformations is also discussed. (J Prosthet Dent 2003;89:109-13.)

Large facial deformities can result from treatment for tumors, trauma, burns, and congenital anomalies.[1] Vascular malformations are rare congenital anomalies. They have a propensity to form in the head and neck region; most of these occur in the skin or scalp.[2] Approximately 35% will involve bone, either directly as an intraosseous lesion, or indirectly because of local expansion.[3,4]

A vascular malformation is defined as a diffuse lesion of dysplastic blood vessels without endothelial proliferation.[3] They are further classified according to the type of vessels involved: capillary, venous, lymphatic, arteriovenous, or other combinations.[2,5]

Arteriovenous malformations (AVM) in the orofacial region are infrequent, with an incidence of 0.8%.[3] They can be congenital or acquired. Rarely, a small congenital malformation can be exacerbated by a traumatic event and become fulminant.[2,6] They typically do not spontaneously regress and can be multifocal. Acquired AVMs can result from injury, surgery, or hormonal changes, such as puberty or pregnancy. These malformations expand by a change in hemodynamics rather than cellular proliferation. Regular blood flow becomes a series of low-resistance shunts, which promotes collateral formation and dilation of abnormal and normal vasculature.[7]

The clinical presentation of an arteriovenous malformation includes a pulsatile mass exhibiting thrills and bruits, an elevated temperature, and dermal staining of the involved overlying skin.[6,7] They are often asymptomatic until expansion results in facial asymmetry, tooth mobility, bony destruction, bleeding, or an obstructed airway.[7,8] Severe pain often accompanies the progression of the disease.

Diagnosis of AVM made on clinical presentation and histologic evaluation alone can be difficult, because the histologic study does not show the hemodynamics of the lesion. Combining multiple imaging techniques is useful in making a more definitive diagnosis. Magnetic resonance imaging and computed tomography are useful in determining the expansion within tissue planes, can define the spatial relationship, and elucidate destructive bone changes or flow voids.[4,7] Doppler flow imaging and angiography can be useful in determining the rhealogic properties of the lesion and distinguishing between low-flow lesions, such as capillary and lymphatic versus high-flow lesions, such as arteriovenous malformations.[3,4,9] Currently, a definitive diagnosis is achieved by evaluation of the clinical, histologic, and radiographic findings by a multidisciplinary team experienced in vascular anomalies.

The primary reasons to treat AVM are to reduce intractable pain, improve function and improve cosmesis.[2] However, complete resection is often impossible to achieve when multiple foci of the lesion exist and may result in more severe facial disfigurement and functional impairment than was being caused by the disease. Treatment of AVM generally consists of embolization therapy (use of occlusive materials to reduce blood flow), followed by surgical resection.[9-11] Intraoperative management is complicated because of the potentially high risk of severe hemorrhage or death by exsanguination.[8] Further, appropriate wound healing may be compromised because of local ischemia, resulting in tissue necrosis. Surgical removal of the necrotic tissue can perpetuate the ischemia-necrosis cycle because of the already compromised blood supply.

Embolization therapy without resection has also been recommended.[3,11] Although this technique may allow preservation of tissue, the result is often short-lived. Large lesions may involve many regional vessels, such that complete occlusion is not technically feasible. Furthermore, embolization can result in rapid re-formation of collateral vessles,[10] thereby contributing to the progressiveness of the disease process.

Therapeutic alternatives include less invasive procedures such as the use of sclerotherapy, cryosurgery, brachytherapy, and partial resection.[9-11] Sclerotherapy is

[a]Associate Dental Specialist and Director of Maxillofacial Prosthetic Services, Department of Restorative Sciences.

the injection of sclerosing agents such as 95% to 100% ethyl alcohol or 3% sodium tetradecyl sulfate.[11] The aim is to cause fibrosis of the vessels, which will reduce blood flow of the arteriovenous malformation. This is not a permanent solution, because it has to be repeated. Cryosurgery, brachytherapy, and partial resections are also not considered to be permanent solutions, but rather conservative management techniques designed to postpone or circumvent the more disfiguring alternatives.

It is clear that all treatment options for AVM may produce results that are refractory and unpredictable. Treatment alternatives do not often provide a defect with clean margins free of disease progression. Surgical reconstruction may not be possible because of a compromised blood supply. Prosthetic rehabilitation may be the only means of addressing esthetic concerns, but is not without limitations. Prostheses have to be remade frequently to allow for changes in tissue contours and erythematous margins. Conventional prosthetic design may have to be modified to provide adequate retention of the restoration. This clinical report describes the maxillofacial prosthetic management of a patient who underwent surgical resection of an arteriovenous malformation of the face.

CLINICAL REPORT

A 42-year-old man was referred by his plastic and reconstructive surgeon to the Maxillofacial Prosthetic Clinic at the University of Minnesota for evaluation. He had been treated over the previous 8 years for a persistent and progressive AVM of the right face, head, and neck area. Numerous surgical excisions resulted in removal of the right ear, the right nasal ala, external malar skin, and the upper and lower lip to the midline. The recent ear surgery had been reconstructed with a scapular pedicle flap. Partial dehiscence of the scapular flap resulted in a large, gaping, weeping wound in the auricular/mastoid region. There were no plans to fabricate an auricular prosthesis until there was complete wound healing of the area. No attempts had been made to surgically reconstruct the face. The patient reported profound, intermittent xerostomia from the cholinergic action of his pain medication, yet he experienced continual drooling because of lack of oral seal.

A comprehensive clinical and radiographic examination was completed. Facial edema, bruits, and warm, erythematous skin indicated progressive disease into the infraorbital region. Radiographic reports indicated the presence of a small intracranial AVM located near the circle of Willis, and localized progression of the superficial facial AVM. No further surgeries were planned at this time. Several teeth demonstrated decay, for which routine amalgam restorations were placed. Custom fluoride carriers were fabricated for use with 0.4% stannous fluoride to prevent further decay.[12] Samples of Biotene mouthwash and Oral Balance moisturizing gel (LaClede Inc, Gardena, Calif.) were given to the patient for oral lubrication and increased oral comfort.

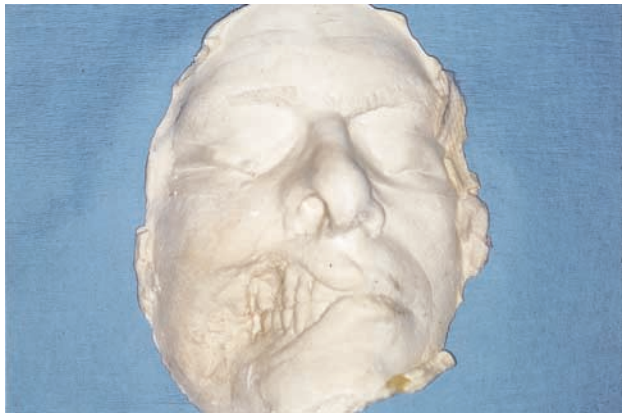

Fig. 1. Complete facial moulage for preliminary design of 2-piece prosthesis.

The patient desired a prosthetic solution for esthetics only. He was made aware of the functional limitations of the prosthesis with regard to speech, mastication, and deglutition. A complete facial moulage was obtained by use of irreversible hydrocolloid (Surgident Orthogel; Miles Inc, South Bend, Ind.), gauze strips (NuGauze; Johnson & Johnson, Arlington, Tex.), and plaster (Modern Materials; Heraeus Kulzer, South Bend, Ind.) for fabrication of a facial prosthesis (Fig. 1).

ERA attachments (Sterngold-ImplaMed, Attleboro, Mass.) were bonded directly on the facial surfaces of the maxillary canine and first molar, and the mandibular canine and first molar, with resin-bonded cement (Panavia 21; J Morita USA Inc, Irvine, Calif.), as previously described[13] (Fig. 2). A vinyl polysiloxane impression (Examix NDS; GC America, Alsip, Ill.) was made of the attachments for fabrication of resin housing units for the matrix portion of the ERA attachments. Heat-polymerizing resin (Lucitone Clear; Dentsply Int, York, Pa.) was used to fabricate the housing units (Fig. 3).

The matrix portion of the ERA attachments was transferred chair side with autopolymerizing resin (Dentsply Repair Material; Dentsply Int.) (Fig. 4). A regional moulage was made with vinyl polysiloxane (Examix NDS; GC America) with the resin matrixes in place on the teeth. Because the defect involved both the upper and lower lips, a 2-piece prosthesis was designed to allow for movement during speech. The wax-up of both facial prostheses were refined and evaluated on the patient. Margin extensions were refined and the wax-up completed. The lower lip wax pattern was sealed to the master cast (Fig. 5). Another impression was made of this master cast and the lower lip wax pattern to capture the position and anatomy of the lower lip, and the lower lip prosthesis. The upper lip/nasal wax-up was sealed on this second master cast; it was designed to overlap the

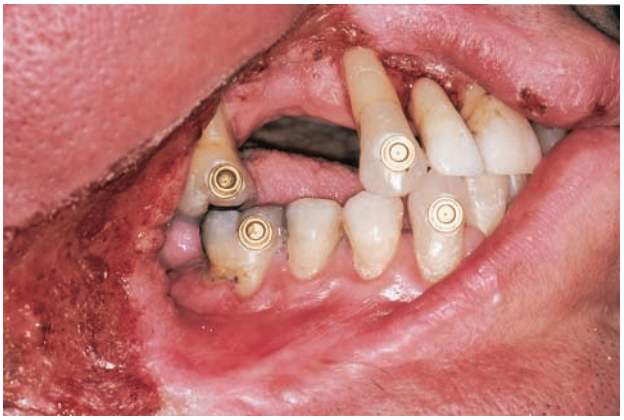

Fig. 2. ERA attachments bonded directly on teeth for use in retaining prosthesis.

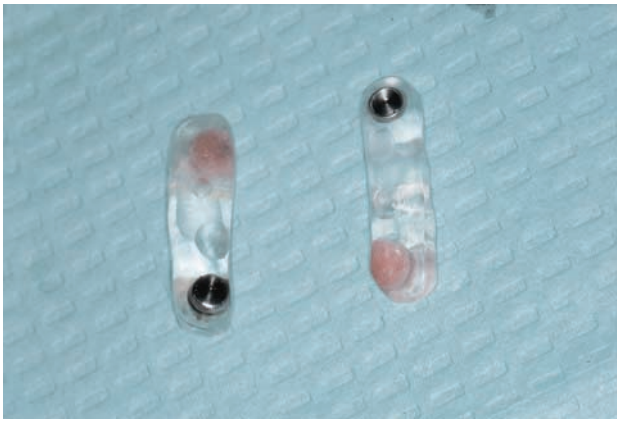

Fig. 3. Clear heat-polymerizing resin used to make housing units for matrix portion of attachments.

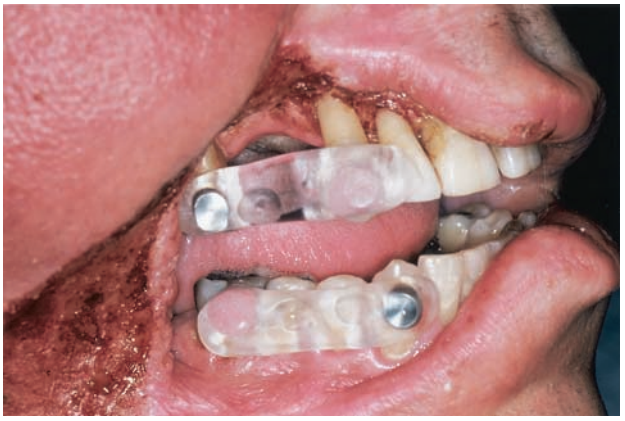

Fig. 4. Matrix portion of attachments transferred and secured with autopolymerizing resin.

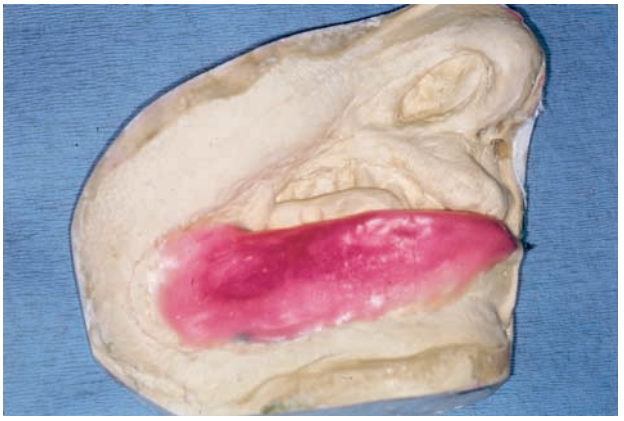

Fig. 5. Wax-up of lower lip is sealed to master cast.

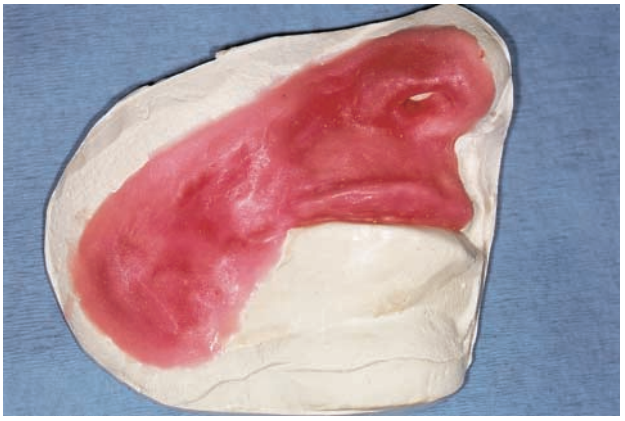

Fig. 6. Transfer impression of wax-up of lower lip to serve as second master cast for processing of upper lip/nose. Wax-up of upper lip/nose prosthesis sealed to master cast.

lower portion of the prosthesis to form a "slip-joint" (Fig. 6).

Each wax pattern and master cast was invested in American Dental Association (ADA) type V dental stone (Die Keen, Heraeous Kulzer) with flasks constructed from 8-inch-diameter commercial grade PVC pipe and processed separately. MDX4-4210 medical grade elastomer (Dow Corning Corp, Midland, Mich.) and Grumbacher oil pigments (Grumbacher Inc, Bloomsbury, Mich.) were mixed to obtain the correct intrinsic color match for the patient. Obtaining a true shade for the skin was difficult because of the erythema surrounding the defect. The patient requested that we attempt to match the true color of his skin, rather than the erythematous tissue. Medical Adhesive type A (Dow Corning Corp) was added to the base component of MDX4-4210 to create a modified cross-linked silicone 2-part prosthesis.[14,15] After the prostheses were completed, extrinsic colorization was completed chair side, and the resin matrixes were bonded to the silicone prosthesis by use of Primer 1205 (Dow Corning Corp) and Medical Adhesive type A. The patient returned for insertion of the prosthesis. The lower lip prosthesis was applied first (Fig. 7), followed by application of the upper lip/nasal prosthesis (Fig. 8). This 2-piece design created a "slip-joint" union at the corner of the mouth to allow for movement of the upper prosthesis over the lower prosthesis during speech. Instructions were given to the pa-

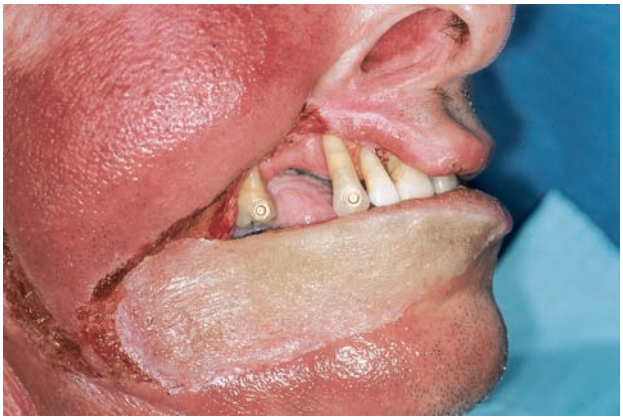

Fig. 7. Lower lip prosthesis in position.

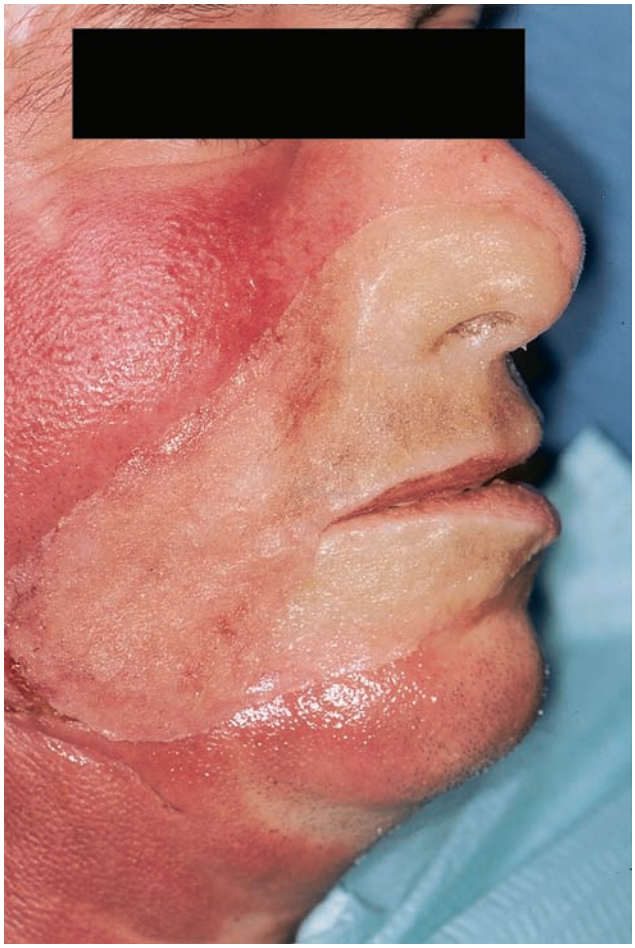

Fig. 8. Upper lip/nose prosthesis in position. Prosthesis overlaps lower lip to allow slippage during articulation.

tient regarding application, removal, and care of the prostheses.

DISCUSSION

The final result after prosthetic treatment was reasonable from a cosmetic standpoint but did not address the functional impairment; drooling continued to be a problem for the patient. It was not possible to achieve a complete oral seal, and saliva slowly escaped from the inferior border of the prosthesis. Further, the margins of the prosthesis were in constant contact with moisture, causing local irritation. However, the patient has been able to wear the prosthesis for brief public appearances.

In this clinical situation, the unpredictable nature of the disease process, localized erythema and edema, and the presence of moisture from the oral cavity necessitated modification of techniques traditionally used for retention of facial prostheses. Although use of the teeth provided improved retention of the prosthesis, this precluded any improvement in masticatory function. However, the patient decided that cosmesis was more important for him because he was being fed via a gastric feeding tube indefinitely.

Although surgical reconstruction of this type of facial defect may have resulted in completion of the oral seal, the cosmetic result would have been marginal because of difficulty in reconstructing the detailed contours of the nasal and mouth areas. In addition, in the progressive course of this disease process, surgical reconstruction poses a high risk for failure and even death. Continued research for methods and materials used in prosthetic reconstruction is needed for these patients for whom surgical reconstruction is not an option.

The difficulty in maxillofacial rehabilitation of large facial defects often involves the compromise of cosmetic versus functional adequacy. The patient can be the only one to determine which aspects of the prosthesis contribute to his/her quality of life.

SUMMARY

This clinical report described the maxillofacial rehabilitation of a large facial defect resulting from surgical excision of an arteriovenous malformation.

REFERENCES

1. Beumer J, Curtis TA, Marunick MT. Maxillofacial Rehabilitation. Prosthodontic and Surgical Considerations. St. Louis: Medico Dental Media International; 1996. p. 377-85.
2. Kennedy KS. Arteriovenous malformation of the maxilla. Head Neck 1990;12:512-5.
3. Prochazkova L, Machalka M, Prochazka J, Tecl F, Klimovic M. Arteriovenous malformations of the orofacial area. Acta Chir Plast 2000;42:55-9.
4. Watzinger F, Gossweiner S, Wagner A, Richling B, Millesi-Schobel G, Hollmann K. Extensive facial vascular malformations and haemangiomas: a review of the literature and case reports. J Craniomaxillofac Surg 1997; 25:335-43.
5. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg 1982;69:412-22.
6. Darlow LD, Murphy JB, Berrios RJ, Park Y, Feldman RS. Arteriovenous malformation of the maxillary sinus: an unusual clinical presentation. Oral Surg Oral Med Oral Pathol 1988;66:21-3.
7. Mulliken JB, Young AE. Vascular birthmarks: hemangiomas and malformations. Philadelphia: WB Saunders; 1988. p. 324-38.
8. Motamedi MH, Behnia H, Motamedi MR. Surgical technique for the treatment of high-flow arteriovenous malformations of the mandible. J Craniomaxillofac Surg 2000;28:238-42.

9. Resnick SA, Russell EJ, Hanson DH, Pecaro BC. Embolization of a life-threatening mandibular vasular malformation by direct percutaneous transmandibular puncture. Head Neck 1992;14:372-9.
10. Berenguer B, Burrows PE, Zurakowski D, Mulliken JB. Sclerotherapy of craniofacial venous malformations: complications and results. Plast Reconstr Surg 1999;104:1-11.
11. Sadove AM, Eppley BL, Gossweiler K, Hathaway R. Mandibular vascular malformations: treatment and long-term results. J Craniofac Surg 1994;5: 289-94.
12. Keene HJ, Fleming TJ. Prevalence of caries-associated microflora after radiotherapy in patients with cancer of the head and neck. Oral Surg Oral Med Oral Patholog 1987;64:421-6.
13. Cheng AC, Morrison D, Maxymiw WG, Archibald D. 1997 Judson C. Hickey Scientific Writing Awards. Lip prosthesis retained with resin-bonded retentive elements as an option for restoration of labial defects: a clinical report. J Prosthet Dent 1998;80:143-7.
14. Udagama A, Drane JB. Use of medical-grade methyl triacetoxy silane crosslinked silicone for facial prostheses. J Prosthet Dent 1982;48:86-8.
15. Farah JW, Robinson JC, Koran A, Craig RG, Hood JA. Properties of a modified cross-linked silicone for maxillofacial prostheses. J Oral Rehabil 1987;14:599-605.

Reprint requests to:
Dr Donna M. Hecker
University of Minnesota
Room 6-284 Moos Tower
515 Delaware St SE
Minneapolis, MN 55455
Fax: 612-624-2660
E-mail: hecke003@tc.umn.edu

0022-3913/2003/$30.00 + 0

doi:10.1067/mpr.2003.23