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Regulating Microvascular Free Flaps Reconstruction in "Schobinger Stage 4" Arteriovenous Malformations of Face

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Abstract

Objectives Arteriovenous malformations (AVMs) are high-flow, aggressive lesions that cause systemic effects and may pose a risk to life. These lesions are difficult to treat as they have a tendency to recur aggressively after excision or embolization. So, it requires a regulating free flap with robust vascular flow averting the postexcisional ischemia-induced collateralization, parasitization, and recruitment of neovessels from the surrounding mesenchyme—a phenomenon precipitating and perpetuating the recurrence of AVM.

Materials and Methods Sixteen patients (12 males and 4 females) with AVMs Schobinger type 4 involving face were treated from March 2015 to March 2021 with various free flaps: three free rectus abdominis flaps, one free radial forearm flap, and twelve free anterolateral thigh flaps were used for reconstruction following the wide local excision of Schobinger type 4 facial AVM. The records of these patients were analyzed retrospectively. The average follow-up period was 18.5 months. The functional and aesthetic outcomes were analyzed with institutional assessment scores.

Results The average size of the flap harvested was 113.43 cm^2. Fourteen patients (87.5%) had good-to-excellent score ($p = 0.035$) with institutional aesthetic and functional assessment system. The remaining two patients (12.5%) had only fair results. There was no recurrence (0%) in the free flap group versus 64% recurrence in the pedicled flap and skin grafting groups ($p = 0.035$).

Conclusion Free flaps with their robust and homogenized blood supply provide a good avenue for void filling and an excellent regulating effect in inhibiting any locoregional recurrences of AVMs

Keywords
- ► facial arteriovenous malformations
- ► microvascular reconstruction
- ► regulating free flaps

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Introduction

The standard of care for large Schobinger stage 4 facial recurrent arteriovenous malformations (AVMs) envisages a multimodality-integrated approach. The successful outcome involves the timely and precise amalgamation of the services of the interventional radiologists (using supra selective co-axial catheters, the perinidus feeders are embolized and the lesion must be devascularized, skeletonized, and deafferented [DSD]), vascular surgeons (wide radical excision with control of bleeding within 48 to 72 hours of DSD) and plastic surgeons (reconstruct the large postexcisional facial defect with compound free flaps). These life-threatening lesions of the face also have many negative implications for the psyche of these patients, with severe compromise on their quality of life. Most of these patients, with their grotesque deformities on the most visible and socially interactive part of their body —the face—with pain, ulceration, crusting, infection, and bleeding episodes—are terribly depressed and frightened. We have reconstructed these stage 4 Schobinger[1] large facial AVMs with compound-free flaps. The reconstruction with large combined free flaps of the postexcisional defect has the following features: 1. With its virtue of homogenized supranormal blood supply, the reconstructed microvascular free flap tissue components suppress the ischemia induced, parasitization, collateralization, and recruitment of microscopic mesodermal vascular residuum to reform arteriovenous fistulas in the bed and thereby the most dreadful complication—the recurrence is prevented.[1,2] 2. The void created by the wide radical excision like the exenterated orbits is effectively filled by the muscle components of the free flap. 3. They also simultaneously address the functional and aesthetic components of reconstruction. 4. Their psychological concerns are effectively addressed by the reconstruction with staged secondary procedures. This study evaluates our algorithmic approach.

Aim

1. To study the effect of a homogenously well-vascularized free flap on preventing the recurrence and relapse of stage 4 Schobinger facial AVM after excision (The regulating effect of free flap).
2. To evaluate our choice of penumbral feeder vessels as recipient vessel during free flap anastomosis.
3. To study the percentage of patients needing secondary procedures following reconstruction with free flaps.

Materials and Methods

Our institutional ethical committee approved the conduction of this retrospective cohort clinical study. Written informed consent was obtained from all patients regarding the use and display of clinical materials, photographs, and videos for research and publication purposes. The study was conducted from March 2015 to March 2021 and included 16 patients (12 males and 4 females) with stage 4 Schobinger facial AVM. All the patients had associated systemic effects of AVM.

Selection Criteria

1. All patients with stage 4 Schobinger facial AVMs.
2. Those patients with large facial defects following wide excision of AVMs reconstructed with microvascular free flaps.

Exclusion Criteria

1. Those patients who were unable to come for the follow-up.
2. Those who had concurrent intracranial AVM extension.

Preoperative Strategies

The plastic surgeon serves as a key member who orchestrates the whole management. The goal of treatment in stage 4 facial AVM is to excise macroscopically and completely all the extracranial malformations with control of all the feeding vessels. For the stage 4 AVM, preoperative DSD using a co-axial supraselective catheterization technique and embolization of all possible perinidus feeding vessels were attempted. All these patients have an element of chronic disseminated intravascular coagulopathy (DIC) with consumption coagulopathy. So, hematologists play a vital role in bringing the coagulation profile to optimum and anemia correction (with fresh whole blood, platelets, fresh-frozen plasma transfusions, vitamin K, steroids, epsilon aminocaproic acid). Cardiologists optimize the valvular and ventricular functions and correct the arrhythmogenic status that is common in stage 4 AVMs. Ulcers, which are also common in this stage 4 AVMs, serve as a portal for overt and subclinical infections. So, culture- and sensitivity-directed antibiotic therapy for 10 to 15 days prior to surgery is commonly practiced.

Operative Strategies

- The vascular surgeon takes control of the most proximal distributing vessel, as in all these cases, ipsilateral or contralateral external carotid artery (ECA).
- A combined plastic and vascular surgery approach to determine the perimeter of the incision taking into account the hairline, ear, eye brow, nose, angle of mouth (the extent of distortion and involvement by the disease).
- Surgery is done in multiple small steps, securing hemostasis at every step. The plane of dissection is kept away from ectatic vessels, and the deep plane of dissection is determined by preoperative imaging, clinical methods, and further refined by intraoperative findings.
- Feeding vessels that were embolized were ligated first as a part of perimeter control. The plastic surgery team prepared the recipient vessels after assessing their quality.

Postoperative Management

Invasive monitoring by the combined effort of physicians and anesthesiologists closely assessed volume shifts. Microvascular flaps were monitored in a standard way. Patients were placed on pressor support in the immediate

Table 1 Institutional aesthetic and functional assessment score

No	Parameters
I	Lagophthalmos in ectropion defect
II	Drooping of angle of mouth and Incontinence
III	Asymmetry of face
IV	Radial expansion and drooping of facial tissues
Scores	
1	Absent
2	Mild
3	Moderate
4	Severe
	Minimum = 4; Maximum = 16
Computation of grades:	
Grade	**Score**
Poor	12–16
Fair to good	8–12
Good to excellent	4–8

postoperative period in all cases. Heparinization was not done in any of the cases. Persistent hypertension was rarely encountered ($n = 1$) in the postoperative period and was taken care of by a physician and cardiologist. Early ambulation was done after 48 hours. All the patients were screened at 6 months, 18 months, and 24 months with high-resolution ultrasonography of the entire reconstructed area and penumbra to detect any locoregional recurrence apart from clinical examination. Two independent observers using institutional aesthetic and functional assessment scores at the end of a 12-month follow-up period assessed the patient's final appearance and functions (►**Table 1**).

Our protocol is illustrated in ►**Fig. 1**

Case Illustration 1

A 38-year-old male with complaints of swelling in the left side of the face for the last 8 years of age, with an increase in size for 2 months. He had uncontrolled bleeding from one side of the lesion for which left ECA ligation was done. The patient presented with dyspnea at rest and bilateral pitting pedal edema for the past 6 months and diffuse soft, compressible, pulsatile swelling of size 18 × 10 cm with thrill and bruit with extensive hemifacial involvement (►**Fig. 2A**). Digitial Subtraction Angiography (DSA) showed AVM in the left side of the face with the cross-circulation with bilateral ECA and recruitment of feeders from the right ECA and left vertebral artery (►**Fig. 2B** and **C**). The left-side ECA was ligated with an acute cutoff with a delayed filling of distal branches from the opposite side (►**Fig. 2B** and **C**). Percutaneous injection of cyanoacrylate glue-tripidol mixture into the AVM was tried, and the patient developed severe facial edema. Hence, further procedures were deferred and excision was done within 72 hours. Whole of left hemiface: skin, subcutaneous tissue, all the superficial mimetic muscles down to the level of the deep facial fascia were excised, leaving behind deep mimetic muscles and facial nerve

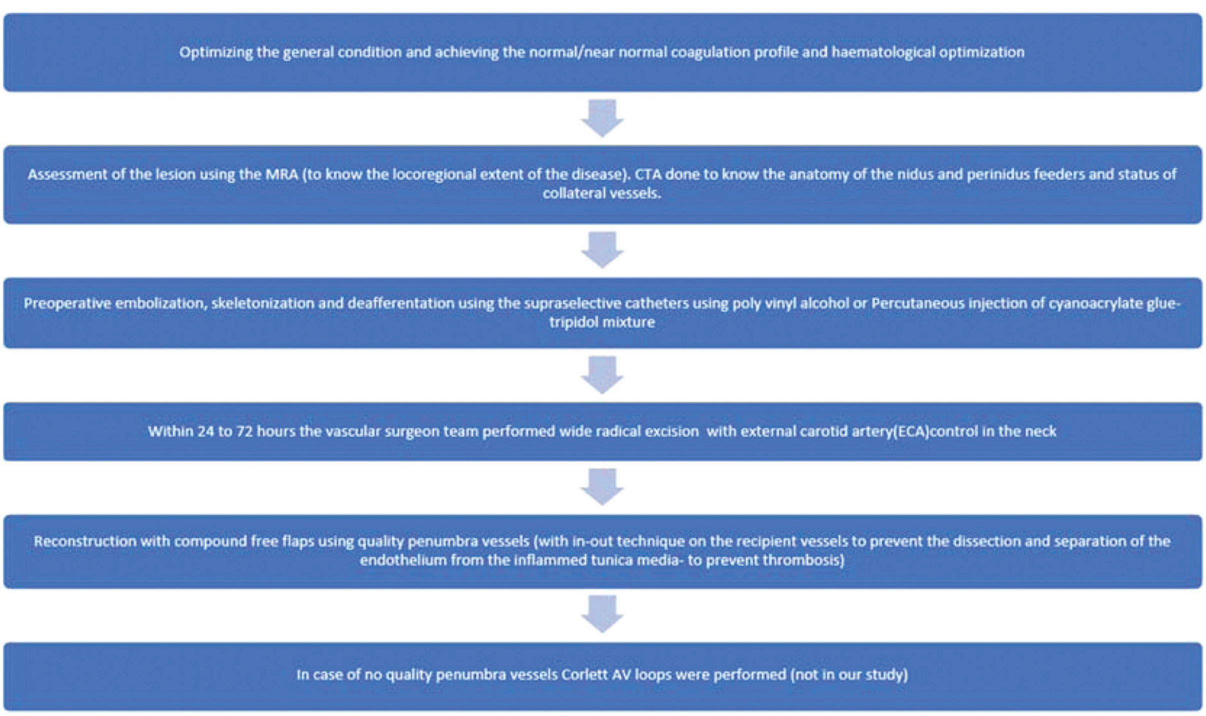

Fig. 1 Our protocol in management. AV, arteriovenous; CTA, computed tomography angiography; MRA, magnetic resonance angiogram.

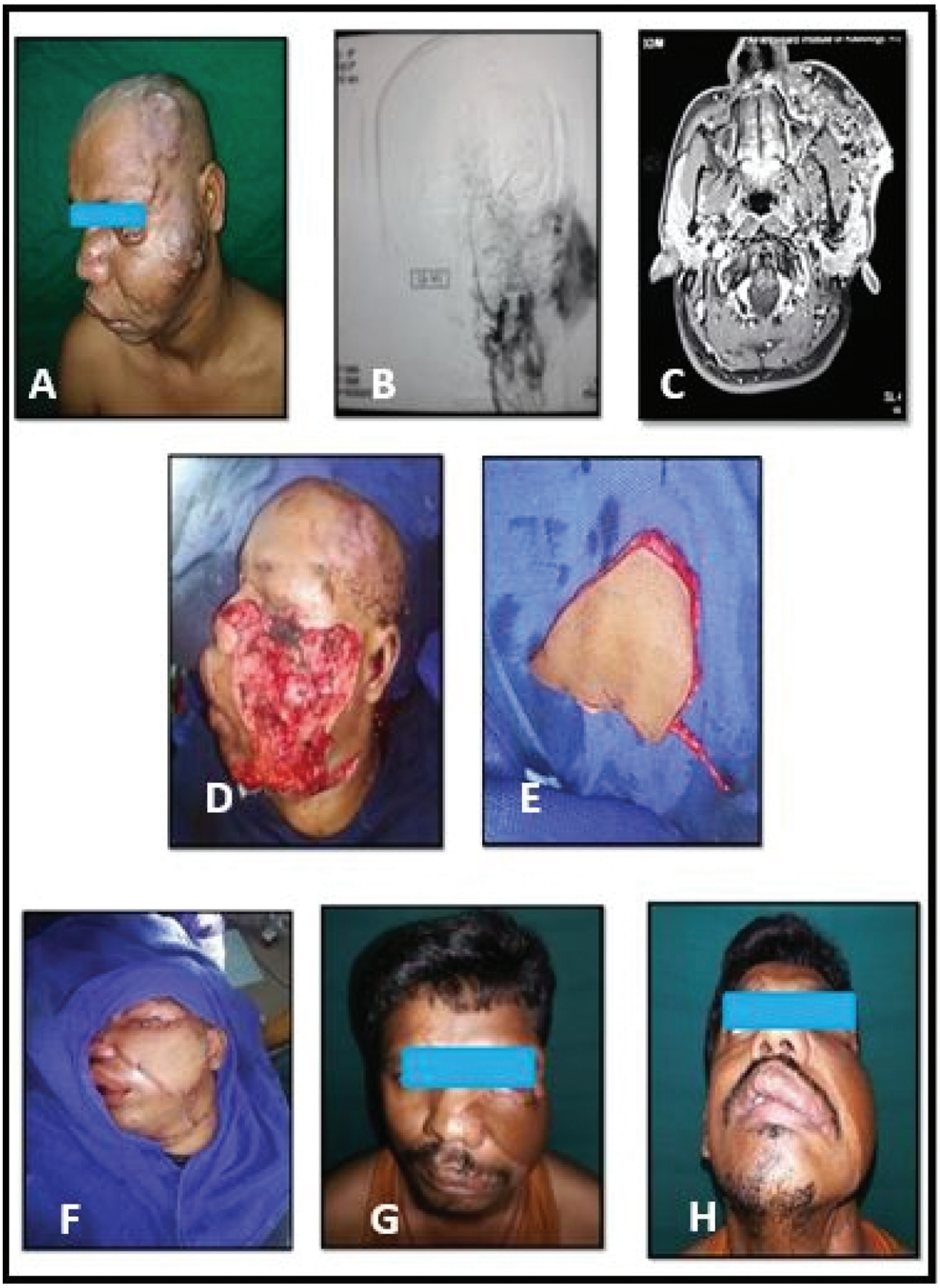

Fig. 2 (**A**) Preoperative picture of case 1. (**B**) Catheter angiogram showing the lesion with nidus. (**C**) Magnetic resonance angiogram pictures of case 1 showing multiple ipsilateral and contralateral feeders and the nidus. (**D**) Postexcisional defect of case 1. (**E**) Harvested anterolateral thigh free flap. (**F**) Immediate postoperative picture of case 1. (**G and H**) Late postoperative picture of case 1.

branches. There was a resultant 16 × 10 × 2 cm post excisional defect over the left hemiface (►**Fig. 2 D**). The free anterolateral thigh flap was designed and harvested from the right thigh (►**Fig. 2E**). The left facial artery and facial vein were identified, dissected, and prepared. The descending branch of Lateral Circumflex Femoral Artery (LCFA) and its venae commitantes were anastomosed with the left facial artery (end to side) and facial vein (end to end), respectively (despite the ligation of ipsilateral external carotid artery the cross over circulation was adequate to produce the pulsatile flow; ►**Fig. 2F**). The flap settled and the patient was followed up for 36 months without any recurrence (►**Fig. 2G** and **H**; ►**Table 2**; case 3).

Case Illustration 2

A 52-year-old male presented with complaints of a large painless mass involving the left eye and left cheek for 10 years. The patient had multiple episodes of profuse bleeding from

Table 2 Demographic details of 16 patients who had undergone facial reconstruction

Case no	Age	Sex	Distribution of AVM on face	Average size of defect postexcision (cm)	Average dimension of the free flap (cm)	Composition and name of the free flap	Complications	Pre-operative morbidities associated	Recipient feeder vessels	Follow-up period (months)	Institution aesthetic and functional assessment score
1	36	M	Left Ch + UL + IO	10*8	12*9	Myofasciocutaneous and right ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	15	5
2	46	M	Right IO + T + O+ F	11*6	12*7	Myofasciocutaneous and left ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	12	4
3	38	M	Left M + UL + Ch + IO + F + T	16*10	18*10	Myofasciocutaneous and right ALT free flap	Uneventful	CCF; U; B; CC; DIC;	Facial artery and vein	36	6
4	52	F	Right SO + T	8*6	10*8	Myofasciocutaneous and transverse rectus abdominis free flap	Uneventful	U; B; CC; DIC	ST artery and vein	20	4
5	56	M	Left SO + T + F	10*7	11*8	Myofasciocutaneous and Left ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	18	4
6	49	F	Right O + IO+ Ch	12*8	13*9	Myofasciocutaneous and left ALT free flap	Lower eyelid ectropion	U; B; CC; DIC	ST artery and vein	18	6
7	37	M	Left O+ IO+ SO+ Ch + T	12*6	13*8	Myofasciocutaneous and right ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	16	4
8	49	M	Right O + T + SO+ F	14*8	15*9	M`yofasciocutaneous and left ALT free flap	Lower eyelid ectropion	U; B; CC; DIC ; CCF	ST artery and vein	20	6
9	28	M	Right M+ Ch + LL + UL	12*5	13*6	Myofasciocutaneous and left ALT free flap	Venous congestion	U; B; CC; DIC	Facial artery and vein	16	5
10	40	M	Left O+ IO+ Ch +T	10*8	11*9	Myofasciocutaneous and transverse rectus abdominis free flap	Lower eyelid ectropion	U; B; CC; DIC ; CCF	ST artery and vein	18	6
11	46	M	Right IO + O+ T + F	16*5	17*6	Myofasciocutaneous and left ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	18	4
12	38	F	Right Ch + UL + IO + T	18*7	19*8	Myofasciocutaneous and left ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	12	4
13	41	F	Left O + IO + Ch	16*6	18*6	Myofasciocutaneous and right ALT free flap	Lower eyelid ectropion	U; B; CC; DIC	ST artery and vein	13	6
14	52	M	Left O + IO + SO	12*6	14*8	Myofasciocutaneous and transverse rectus abdominis free flap	Uneventful	U; B; CC; DIC; B/L PKD; CCF;	ST artery and vein	28	4
15	35	M	Left T + IO + O + UL + Ch	18*7	20*8	Myofasciocutaneous and right ALT free flap	Uneventful	U; B; CC; DIC	ST artery and vein	12	4
16	19	F	B/L SO+ F + Ant S	18*8.5	19*10	Fasciocutaneous and free left radial forearm flap	Uneventful	B; CC; DIC;	ST artery and vein	24	4

Abbreviations: ALT, anterolateral thigh; Ant S, anterior scalp; B, bleeding; B/L PKD, bilateral polycystic kidney disease; B/L, bilateral; CC, consumptive coagulopathy; CCF, congestive cardiac failure; Ch, cheek; DIC, disseminated intravascular coagulopathy; F, forehead; IO, infra orbit; LL, lower lip; O, orbit; SO, supra orbit; ST, superficial temporal; T, temporal; U, ulceration; UL, upper lip.

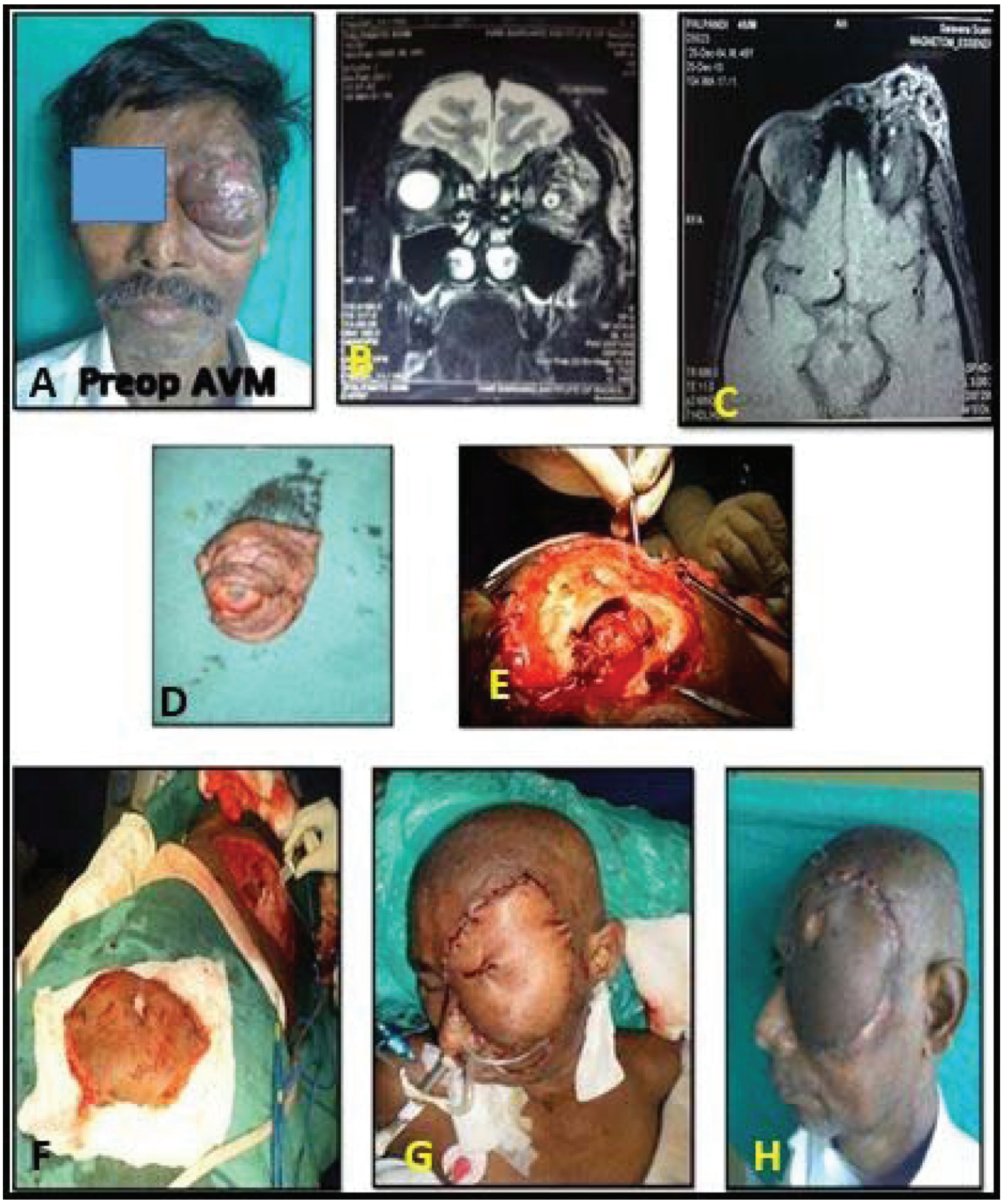

Fig. 3 (**A**) Pre operative picture of case 2. (**B** and **C**) Magnetic resonance angiogram showing arteriovenous malformations causing phthisical bulbi. (**D**) Specimen postexcision of case 2. (**E**) Postexcisional defect of case 2. (**F**) Harvested free transverse rectus abdominis myocutaneous flap of case 2. (**G**) Immediate postoperative picture of case 2. (**H**) Late postoperative picture of case 2.

the upper eyelid and decreased vision for a period of 1 month (►**Fig. 3A**). On examination, the patient had pallor, high blood pressure, bilateral palpable kidneys (bilateral polycystic disease), dyspneic at rest, and signs of congestive cardiac failure for 3 months. The patient had a large 10×8 cm pulsatile mass with dilated veins, pulsatile proptosis, corneal scarring, and conjunctival congestion involving the left orbit and frontotemporal region of the scalp. Magnetic resonance angiogram showed AVM involving the extraocular muscles of the left eye had feeders from the left supraorbital artery and superficial temporal artery with no intracranial extension (►**Fig. 3B** and **C**). With no residual vision in the eye, left orbital exenteration and excision of the AVM were done, resulting in a soft tissue defect measuring 12×6 cm involving the left orbit and infraorbital region that was devoid of periosteum (►**Fig. 3D** and **E**). The defect was reconstructed using a free transverse abdominis myocutaneous flap (►**Fig. 3F**.) The deep inferior epigastric artery and its venae commitantes were anastomosed with the superficial temporal artery and vein, respectively. The postoperative period was uneventful (►**Fig. 3G**) and the patient was followed up for 28 months with no evidence of recurrence (►**Fig. 3H**; ►**Table 2**; case 14).

Case Illustration 3

A 19-year-old girl (►**Fig. 4A** and **B**) presented with 15×8 cm AVM in the forehead region for the past 5 years associated with tachycardia, hyperdynamic circulation, hypertension, and one episode of bleeding in the last 3 months. After magnetic resonance angiogram

Fig. 4 (**A** and **B**) Preoperative pictures of case 3. (**C**) Magnetic resonance angiogram showing large arteriovenous malformations with feeders from both sides. (**D**) Free radial forearm flap of case 3. (**E**) Postexcisional defect of case 3. (**F**) Anastomosis in progress to recipient superficial temporal feeder vessels. (**G** and **H**) Late postoperative picture of case.

(►**Fig. 4C**), she had undergone DSD, with polyvinyl alcohol. She had a hypotensive episode following the DSD; her general condition was stabilized, after which she was taken up for wide excision with ECA control on both sides. Postexcisional defect measuring approximately 18×8.5 cm was reconstructed with primarily thinned free radial forearm flap (►**Fig. 4D** and **E**). The feeder superficial temporal artery and superficial temporal vein were used as recipients (►**Fig. 4F**). She had uneventful postoperative recovery. At 24 months, she had final computed institutional and aesthetic outcome score of good to excellent. There was no recurrence (►**Table 2**; case 16).

Results

The average age of patients in our study was 43 years old. Out of the 16 patients, 12 were male and 4 were female (►**Table 2**). Following were the associated comorbid conditions: chronic DIC ($n=16$) with intermittent pulsatile bleeding ($n=5$), high output cardiac failure ($n=4$), uncontrolled systemic hypertension ($n=4$), diabetes mellitus ($n=2$), and polycystic kidney disease ($n=1$). The average size of the defect after excision of the AVM was 75.18 cm^2. The average size of the flap harvested was 113.43 cm^2. Twelve patients were reconstructed with an anterolateral thigh free flap, one with primarily thinned free radial forearm flap, and three patients with a transverse rectus abdominis free flap. All 16 flaps survived well, while one flap ($n=1$; 6.25%) alone had venous congestion by 48 hours, for which exploration and revision venous anastomosis were done. After that, flap survived uneventfully. Two patients came with late complications of lower lid ectropion by the end of 6 months ($n=2$) (12.25%) for which secondary procedures such as fascia lata sling and blepharoplasty were done. In 14 patients (87.5%), the recipient's vessels were superficial temporal vessels, and in the rest, they were the facial vessels (12.5%). None of them (0%) had any locoregional recurrences at the end of the average follow-up period of 18.5 months. Fifteen patients (93.75%) had pre-excisional DSD. One patient (6.25%) had pre-excision intralesional sclerotherapy. Four patients required (25%) required facia lata sling for drooping angle and blepharoplasty as the secondary adjunct procedure to improve the appearance. Twelve patients (75%) had features of congestive cardiac failure in the preoperative period and had completely recovered in the postoperative period. Thirteen patients (81.25%) had ulceration and bleeding episodes. Sixteen patients (100%) had hematological and coagulation derangements. All these derangements got corrected postoperatively. Fourteen patients (87.5%) had good-to-excellent score ($p=0.035$) with institutional aesthetic and functional assessment system (►**Table 1**). The remaining two patients (12.5%) had only fair results. Retrospectively recurrence rate was compared with the age and sex matched 14 cohorts ($n=6$ pedicled flaps; $n=8$ skin grafting for similar Schobinger stage 4 facial AVMs) by multiple variants regression

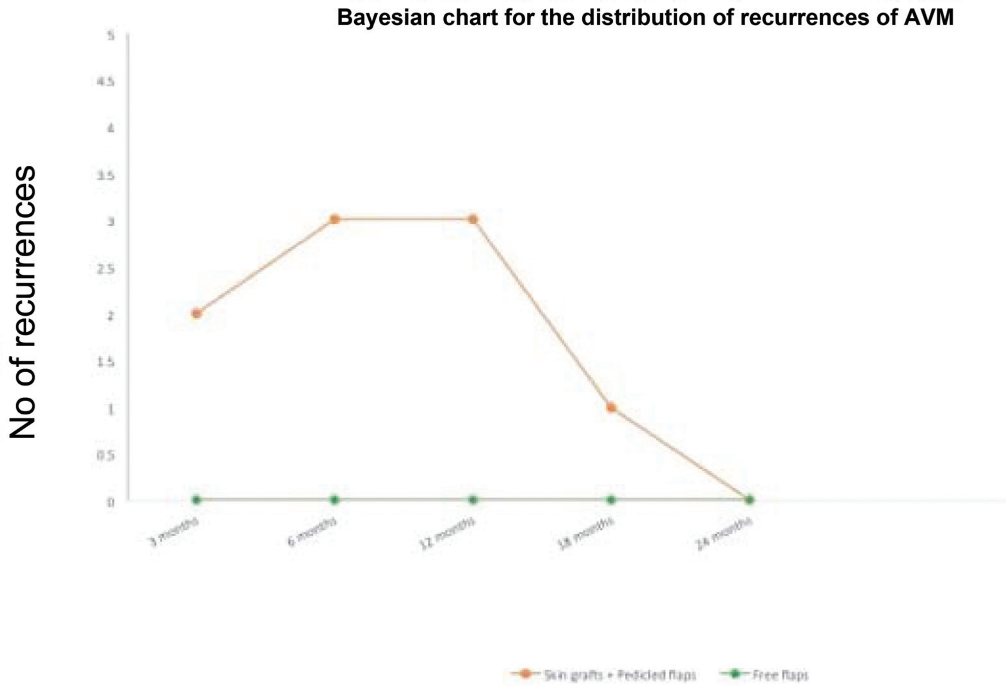

Fig. 5 Bayesian chart showing the distribution and timeline of recurrences in free flaps versus other modality of interventions.

analysis. All these cohorts were operated by the same first author. This showed nil recurrence rate (0%) in the free flaps group versus 64% recurrence rate in the pedicled flaps and skin grafting groups ($p = 0.035$; chi-squared test; ►**Fig. 5**).

Discussion

The aberrations occurring between the 4th and 6th week of intrauterine life lead to the persistence of mesodermal vascular residue in the pharyngeal arches that later in life leads to the development of the AVMs of the head and neck region.[3] The most challenging aspect of the AVMs is the recurrence.[4] The aggressive recurrent nature of AVMs is attributed to the unfavorable hemodynamics within the shunt that diverts blood away from high resistance neighboring tissues rendering them more ischemic.[5–7] Dompmartin et al, in their study, have discerned that introduction of well-vascularized tissue onto the AVM reduced the surrounding ischemia thus preventing the recurrence.[8] Histological changes in the residual AVM after the transfer of free flap on it were studied by Tark and Chung.[9] In this study, histologically thick-walled vessels with abundant elastic fibers underwent a change in the fourth postoperative month with the disappearance of thick-walled vessels leaving behind homogenous thin-walled vessels. No recurrence of the lesion with the disappearance of the existing lesion threw light on the use of free tissue transfer as an important modality in controlling AVMs. This effect is called the "Regulating free flap effect." Free flaps also provide bulk to the facial reconstruction and can provide functional reanimation if combined with muscle flaps.[10] In our study, we have devised an institutional functional and aesthetic score to evaluate patient satisfaction and to know the need for secondary procedures. In this study, the authors used free flaps in the reconstruction of Schobinger facial stage 4 AVM with promising results of no recurrence till the average follow-up period of 18.5 months. The majority of the patients in our study were satisfied with their facial appearance after free flap reconstruction. Keeping in mind, the advantage of using a free flap as a curative treatment modality in stage 4 AVMs, the main challenge faced in free flap transfer is the choice of recipient vessels. Choosing the feeding artery or drainage vein of the AVM as recipient vessels seems to pose a risk for microvascular thrombus formation due to turbulence, existing thrombosis, and consequent damage of tunica media.[1,11–13] Bradley et al[14] concluded that free flaps can be used in the postexcisional defect reconstruction of AVM if the site of microvascular anastomosis was well away from the pathologic ectatic vessels. In contradiction to the previous study, Yamamoto et al in 1994 conducted a case series on 12 patients with facial and extremity AVMs where feeder vessels were used as recipient arteries with no evidence of thrombus formation or occlusion. Adding to this, Tark and Chung[9] in the year 2000 used engorged dilated feeding arteries as recipient vessel in a series of three patients with head and neck AVM successfully. Out of the 22 consecutive patients who underwent free flap transfer after AVM resection studied retrospectively by Fujiki et al[15] et al in 2018, 11 were located in head and neck region with the remaining 11 in extremities. Feeding arteries were used as recipient vessels in the microvascular reconstruction after AVM excision in extremities with no flap failure. Pribaz et al[16] reported total flap necrosis due to venous thrombosis when feeding veins were used as recipient vein. However, no study till date reports arterial thrombosis when feeder vessels were used. We in this study, first of its kind, have used feeder vessels as recipient vessels in this series of 16 patients with facial Schobinger stage 4 AVM with no complications except in one patient with venous thrombosis that was also revised and salvaged. The efficacy of using normal vein as recipient vein in free flap microvascular anastomosis is well established by various studies.[17–19] No literature till date to the best of our knowledge reports the effect of choosing drainage vein as a recipient vessel in head and neck AVMs. Fujiki[15] et al reported successful usage of drainage vein as recipient vein in 11 patients with extremity AVMs. However, they used normal vein for microvascular anastomosis in head and neck AVMs. Few studies discerned the pathophysiology causing drainage veins as an unsuitable choice as recipient veins. The turbulent high-pressure flow in the arterialized veins causes shearing stress within the endothelium leading to medial smooth muscle hyperplasia ultimately ending up in intimal hyperplasia.[11–13] We have used drainage veins as recipient veins in all patients with the careful technique of in-out suturing fashion for recipient veins. This along with diligent handling of vessels adds to the success of the free flap survival in our series. We used the penumbral feeder vessels in all our cases for the following reasons: 1. Proximity. 2. Use of intercalary vein or arterial graft to reach the neck vessel or the desired vessel away from the AVM penumbra zone may result in thrombosis. 3. Our diligent technique of anastomosis in utilizing the feeder vessels as recipient vessel also brings about inhibition of parasitization, collateralization, and recruitment of the mesenchymal vascular tissue in the penumbra leading to recurrence.[17,20] Compared with Hong et al[21] study ours is relatively large study and demonstrated the successful usage of recipient feeder vessels and regulating effect of free flaps preventing the recurrences. Wong et al[22] in their case report mentioned about excision of AVM in the hemiface leaving behind an unresectable macroscopic element around the internal carotid artery and followed for the maximum of 3 years. At the end of follow-up, they had another new lesion in the cheek region but the residual left out lesion remained quiescent with no flare up. In our study, we completely excised macroscopically all the disease dissecting through the uninvolved penumbra region leaving no residual disease. The limitation of our study is the small sample size. However, our study with a structured protocol and prescribed technique of microvascular anastomosis for recipient veins had brought a 100% success rate that envisaged the careful and diligent use of feeder vessels.

Conclusion

The free flaps endowed with the supra-normal homogenized blood supply capable of exerting the regulating effect by completely eschewing the recurrence—the most dreadful complication—was noted in our study. The use of penumbra feeder vessels as recipient vessels with diligent microvascular technique may increase the successful outcome. Well-planned and executed free flaps added the aesthesis to the reconstruction of the face region in a single stage operation.

Conflict of Interest
None declared.

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