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Extensive Facial Arterio-Venous Malformations; Combined Approach, Embolization Followed by Surgical Reconstruction

TAREK SHOUKR, M.D.*; HAZEM MOHAMED ABD EL KHALEK, M.D.** and MOHAMED SAAD SADAKA, M.D.*

The Departments of Plastic & Reconstructive Surgery and Neuropsychiatry**, Faculty of Medicine, Tanta University*

ABSTRACT

Background: Arteriovenous malformations (AVMs) are composed of an abnormal communication between arterial and venous channels without intervening capillary bed. In the face, they commonly present with asymmetry, bleeding, tooth mobility and headache. The most successful treatment to date is embolization followed by surgical excision.

Objectives: To evaluate the safety and efficacy of embolization followed by surgical reconstruction in the treatment of extensive facial AVMs.

Patients and Methods: In this study we present the outcome of treatment of 25 patients presented with extensive facial AVMs and admitted in the department of plastic and reconstructive surgery at Tanta University Hospital in the period from January 2016 to July 2021. Our treatment strategy was embolization guided by digital subtraction angiography followed by surgical excision.

Results: Twelve patients had one anatomic facial subunit affected by the lesion while nine cases had two subunits affected and four patients had 3 subunits affected. The average length of the widest dimension of the lesion was 6cm. Most of our cases (19 of 25) were in Schobinger stage 2 at the initial presentation. All our patients were followed for at least 1 year and none of them showed any evidence of recurrence of the lesion during the follow-up period. All our cases had controlled disease.

Conclusion: Preoperative embolization followed by surgical excision is the ideal treatment for extensive AVMs of the face.

Key Words: *Embolization – Arteriovenous – Malformation – Facial.*

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Ethical Considerations: This research was carried out in accordance with the principles of the Helsinki Declaration. Approval was obtained for this study from the Ethical Committee of Faculty of Medicine, Tanta University. An informed written consent was obtained from all patients regarding surgical procedures and publication of their photos.

Correspondence to: Dr. Mohamed Saad Sadaka,
E-Mail: mohamed.sadakayyy@gmail.com

INTRODUCTION

Mulliken et al., classified the vascular anomalies according to their biological features into two main categories; infantile hemangiomas and vascular malformations which may be capillary, arterial, venous, lymphatic, and combined **[1]**.

The incidence of Arteriovenous malformations (AVMs) is about 0.1% of the general population. The AVM nidus is an abnormal communication between the arterial and venous pathways without intermediate capillaries **[2]**. Arteriovenous malformations still do not have a clear etiology or pathogenesis. The current most possible theory is a vascular development disturbance occurring during weeks 4th to 6th intrauterine **[3]**.

Common presentations of AVMs of the face are asymmetry, bleeding, mobility of teeth, and headache. AVMs are present at birth at the tissue level. Nearly 60% of the cases are diagnosed at birth, while the remainder are diagnosed during childhood or adolescence. The malformation typically increases in size over time **[4]**.

Instead of cellular multiplication, it is thought that their enlargement is the result of increased blood flow. There have been reports of rapid expansion in response to infection, attempted excision, local trauma, ligation, and hormonal changes (puberty or pregnancy). The arterio-venous shunting's "steal phenomenon" reduces the nutrient flow to the skin, which could lead to ischemic necrosis and repeated bleeding **[5]**.

Early treatment is required as their natural course is progression and tissue destruction. For the treatment of head and neck AVMs, a variety of techniques, have been employed over the past ten years such as endovascular embolization, sur-

gical excision, laser therapy, or combination therapy. Embolization, followed by surgical resection, has been shown to have the best success rates in the treatment of AVMs **[6]**.

In this article we present our experience with the treatment of extensive facial AVM using a combined approach; embolization followed by surgical resection and reconstruction.

PATIENTS AND METHODS

This study included 25 cases admitted to Tanta University Hospital with the diagnosis of facial AVM in the period from January 2016 to July 2021. Patients who have undergone any previous form of treatment for their AVM were excluded from the study. All patients underwent detailed history taking and physical examination.

All embolization procedures were performed under general anesthesia with heparinization of 30-50 IU/Kg, femoral puncture with insertion of sheath 6 French, and digital subtraction angiography performed for all head and neck arteries including both external and internal carotid arteries and both vertebral arteries.

Our main feeders usually branched from external carotid arteries according to the location of the AVM including facial, superficial temporal, internal maxillary, occipital and posterior auricular arteries.

Selective catheterization was performed for the feeding arteries with microcatheters according to the available materials in the hospital including marathon, Apollo, Rebar 27 microcatheters (Medtronic, Minnesota, USA) sonic 1.5 (Balt, Montmorency, France). According to the feeders and velocity of the malformation, the embolizing material was decided; either onyx 18 (Medtronic, Minnesota, USA) or polyvinyl alcohol (PVA) like Contour particles 250-350μm (Boston scientific, Massachusetts, USA).

Nidus embolization was performed with most of feeders occluded to devascularize the malformation as much as we can before referring to the patient to surgery.

Surgical resection was performed within 2-5 days (average 3 days) post-embolization. All operations were performed under general anesthesia. Complete resection of the malformation was performed followed by primary closure or reconstruction using grafts or local flaps according to the situation.

RESULTS

This study included 25 patients presented with facial AVM. Their age ranged from 3 to 72 years (average 25 years). Seventeen patients were males and eight were females. The length of the widest dimension of the lesion in our patients was in average of 6cm (range from 2-11cm).

None of our patients required a second embolization session before surgery. After resection of the lesion, the wound could be closed primarily in 13 patients while three patients required skin grafting and nine patients required closure by local flap (Table 1).

Table (1): Demographic and clinical data of our patients.

Variables	Patients' data, number and percentage		
Age	3-72 years (average 25 years)		
Duration of lesion from onset to presentation	2-63 years (average 19 years)		
Length of the largest dimension of the lesion	2-11 cm (average 6 cm)		
Time interval between embolization and surgery	2-5 days (average 3 days)		
Follow-up period	16-28 months (average 22 months)		
Gender	Males: 17 (68%)	Females: 8 (32%)	
Material used for embolization	Onyx 18: 16 (64%)	(PVA) like Contour particles: 9 (36%)	
Schobinger stage at presentation	Stage 1: 3 (12%)	Stage 2: 19 (76%)	Stage 3: 3 (12%)
Number of facial subunits affected by the lesion	1 subunit: 12 (48%)	2 subunits: 9 (36%)	3 subunits: 4 (16%)
Method of reconstruction after resection	Primary closure: 13 (52%)	Skin graft: 3 (12%)	Local flap: 9 (36%)

The postoperative period was uneventful in all cases. No blood transfusion was required in any case, whether preoperative or postoperative. No complications were recorded in any case during the hospitalization period. All cases were discharged few days after surgery.

Only three cases developed minor wound dehiscence that was managed conservatively by wound care and follow-up and healed completely.

All cases were followed for at least 1 year postoperative and no cases showed any clinical evidence of recurrence of the malformation during the follow-up period (Figs. 1-4).

The Wu et al., approach with four kinds of results was used to evaluate our ultimate outcome: (1) Controlled disease, (2) Improved disease (residual, no expansion), (3) Persistent or stable disease (neither worsened nor improved), and (4) Worsened or recurrent disease **[7]**. All our cases fell into the first group (controlled disease). We had no patients of the other 3 categories.

Regarding the number of anatomic subunits of the face that were affected by the lesion; it was one subunit in 12 patients, two subunits in 9 patients and three subunits in 4 patients. The most commonly affected subunit was the cheek. At the initial presentation, three of our patients were in Schobinger stage 1, nineteen patients were in Schobinger stage 2 and three patients were in Schobinger stage 3.

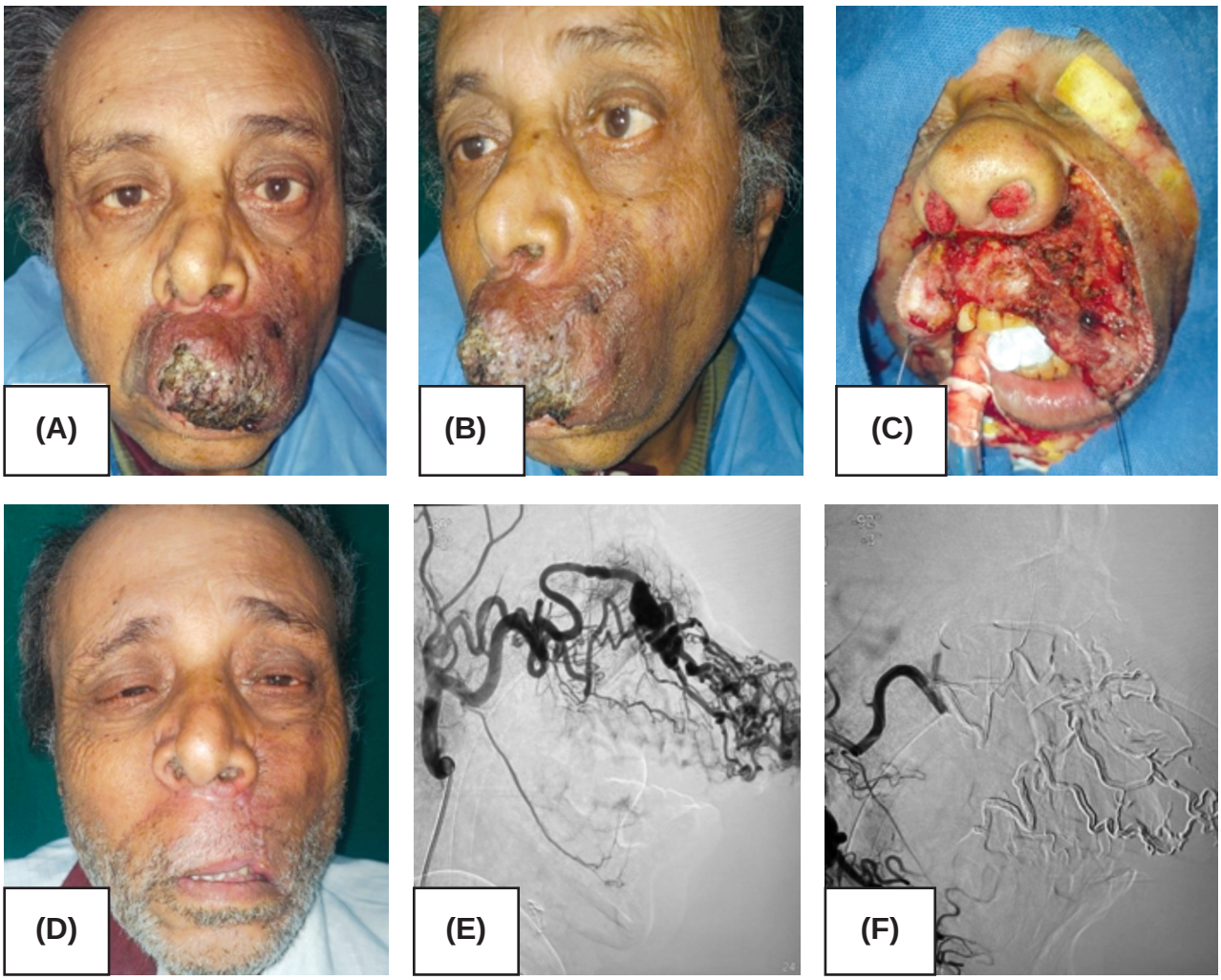

Fig. (1): Sixty-seven years old patient with AVM of upper lip and left cheek. (A): Preoperative frontal view. (B): Preoperative oblique view. (C): Intraoperative view after excision of the lesion. (D): Postoperative view 1 year after operation. (E): Pre-embolization angiographic view. (F): Post-embolization angiographic view.

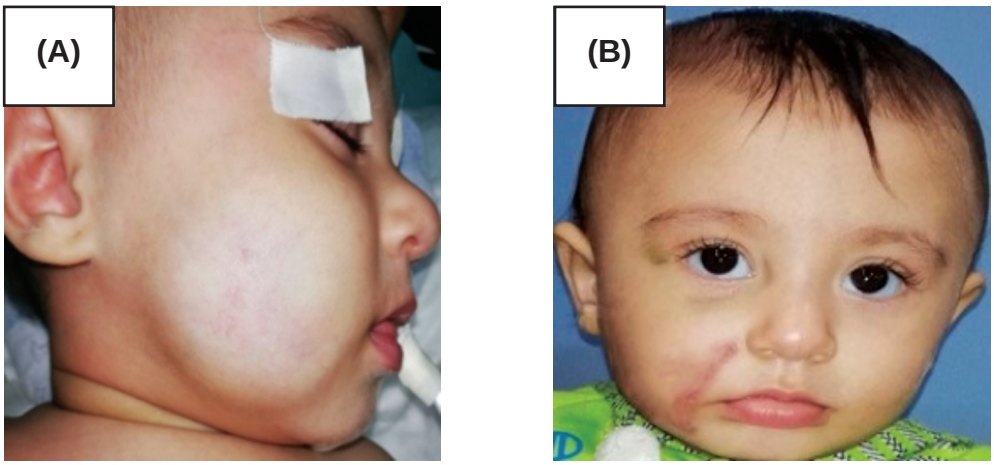

Fig. (2): Three-year-old patient with AVM of right cheek. (A): Preoperative view. (B): Postoperative view.

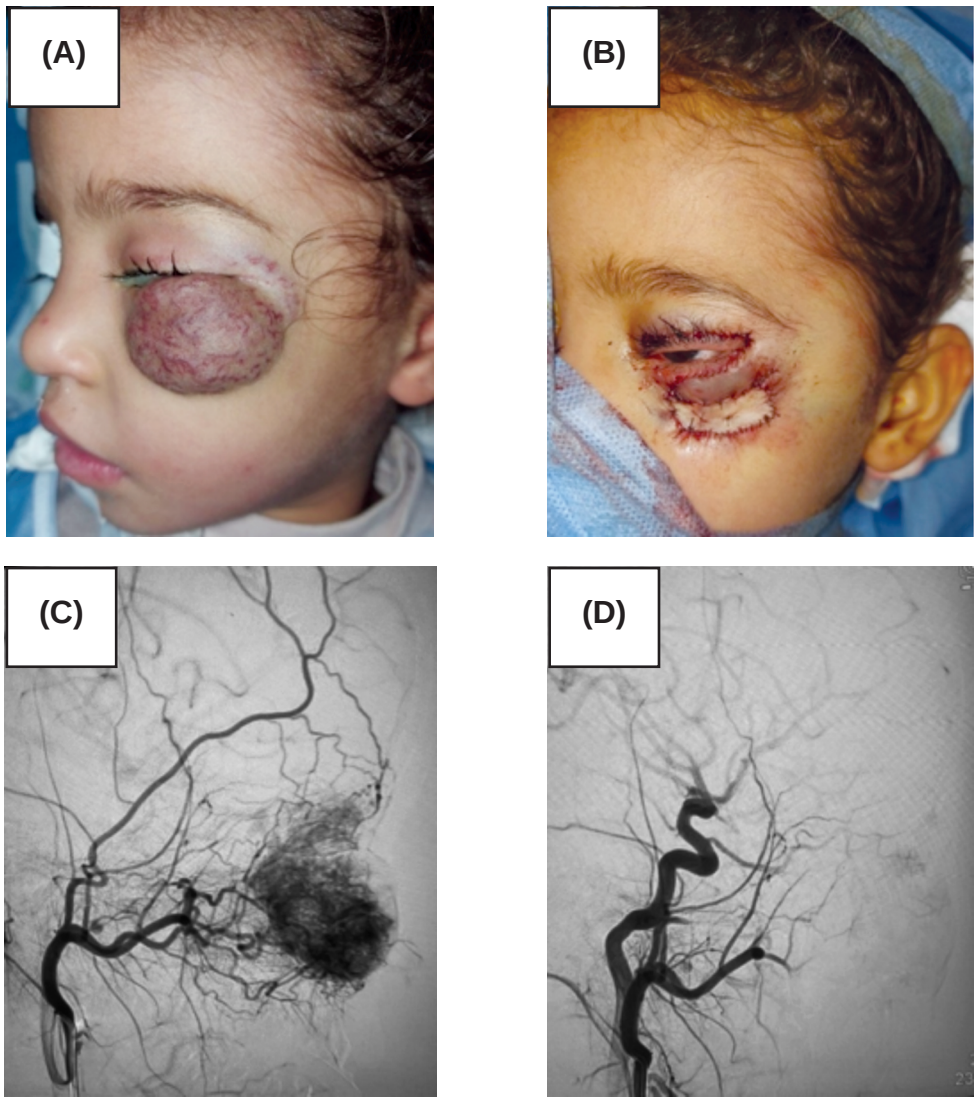

Fig. (3): Six years old patient with AVM of left lower eyelid and left cheek. (A): Preoperative view. (B): Postoperative view. (C): Pre-embolization angiographic view. (D): Post-embolization angiographic view.

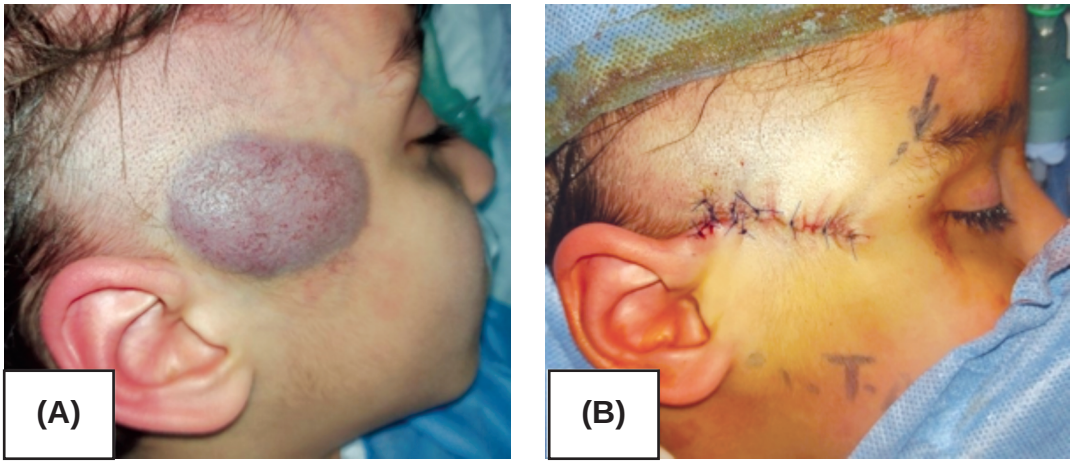

Fig. (4): Five years old patient with AVM of right cheek. (A): Preoperative view. (B): Postoperative view.

DISCUSSION

Before the recent developments in the field of endovascular therapy, the treatment of AVM was mainly through surgical excision or ligation of supplying arteries **[8]**. However, surgical excision alone has several challenges: (i) The risk of bleeding, which can be fatal; (ii) The difficulty in determining the extent of the lesion resulting in extensive surgical resection; (iii) The potential for recurrence beginning from dormant shunts localized away from the central nidus **[9]**.

Transarterial and transvenous embolization has evolved into the ideal adjuvant to surgery with the development of endovascular treatment methods and new embolic agents. The catheterization of feeding arteries near to the nidus has been made possible by recent advancements in the design of micro catheters and distal navigation techniques. Various methods such as liquid glue, coiling, and sclerotic agents can be used when trans-arterial embolization is possible **[10]**.

For embolization, a variety of sclerosing agents can be applied. Onyx 18 is utilized when the catheter probe could be inserted inside the nidus. Particles (Embosphere) of the right size are applied via a more proximal feeding artery for lesions where the catheter could not be inserted within the nidus **[3]**. When ethanol is used, there is considerable complication rate of 10-15%, including skin necrosis, haemoglobinuria, neuropathy and cardiovascular collapse. Complications of glue embolization include skin necrosis, ischemia of the adjacent normal tissue and non-targeted embolization **[11]**.

Direct percutaneous puncture with subsequent embolization can be a viable alternative when there is inadequate vascular access to the vessels supplying the AVM. The end-point of surgery is determined by the extent of the lesion which is sometimes difficult to judge. A more conservative resection will lead to recurrence of the lesion, while an aggressive resection may cause permanent damage to adjacent tissues **[12]**.

All our patients underwent preoperative digital subtraction angiography. The preoperative imaging as angiography defines limits and extension of the nidus, its feeding vessels and flow characteristics **[13]**.

The interval between embolization and surgery in our study was 2 to 5 days (average 3 days). Based on the theory that waiting longer could cause the lesion to expand and grow, the majority of current literature suggest waiting 1 to 3 days between embolization and excision **[14-16]**. However, Pedreira et al., found no significant difference in recurrence rates between patients receiving immediate (1 day) or delayed (10.6 day) resection after embolization, and they concluded that delaying resection, as long as the window remains subacute, may actually have benefits with regards to the preservation of aesthetics and function because there will be less swelling and inflammation in the surgical field, which will lead to increased precision of dissection **[17]**.

Selective embolization will mostly fail if used as the only treatment for high flow lesions because new collaterals develop rapidly. Also, ligation of main arteries supplying AVM is prohibited due to high failure rate and obstructing the pathway for later embolization **[18]**.

According to the outcome evaluation approach described by Wu et al. **[7]**, all our patients fell into

the first group (controlled disease). In their meta-analysis on AVM, Kansy et al., noticed that embolization plus surgery was the most effective treatment method in 41/49 cases (83.7%), followed by embolization alone in 13/20 cases (65.5%), dominant outflow vein sclerosant embolization in 5/14 cases (35.7%), and surgery alone in 4/5 cases (80%). They also found that symptomatic improvement (improved disease) occurred in 6/49 cases (12.2%) using embolization plus surgery, in 7/20 cases (35%) by embolization alone, in 7/14 cases (50%) by dominant outflow vein sclerosant embolization and in 1/5 cases (20%) by surgery alone. Stable disease was achieved in 2/49 cases (4.1%) after combined therapy and in 2/14 cases (14.3%) following sclerosing of the dominant outflow vein. Recurrent or worsened disease was the outcome in only two cases after combined therapy (2/49; 4.1%) **[3]**.

Preoperative embolization facilitates the surgical excision of AVM by reducing the size of lesion and allows an excellent scar placement. It also makes the lesion firmer allowing safer dissection with protection of surrounding vital structures and preserving the volume of facial soft tissues which results in an excellent aesthetic outcome.

Large high flow AVMs were previously considered unresectable and accordingly, patients received only palliative embolization monotherapy with persistent high failure rate mainly due to the extent of the lesion. On the contrary, complex surgical resections for extensive lesions after superselective embolization are now possible with high success rate thanks to the application of modern technologies and appropriate interdisciplinary planning **[14,19]**.

Conclusion:

We can conclude that embolization followed by surgical resection within 2-5 days is a safe and successful strategy in the treatment of extensive facial AVMs with minimal complications profile and no postoperative recurrence.

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