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Original Research Article

Single stage surgical excision for vascular malformation without any pre-operative adjuvant therapy: our experience regarding case selection in 20 patients

Narendra G. Naik[1], Sangram Karandikar[2]*

[1]Department of General Surgery, Rajiv Gandhi Medical College, Kalwa, Thane, Maharashtra, India
[2]Department of General Surgery, Terna Specialty Hospital and Research Centre, Navi Mumbai, Maharashtra, India

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***Correspondence:**
Dr. Sangram Karandikar,
E-mail: sangkaran@hotmail.com

ABSTRACT

Background: Vascular malformations generally present in young population. The patients or their guardians seek treatment for alleviation of functional and/or cosmetic disfigurement. Non-invasive techniques with or without surgery are available, however surgical treatment has been found to be most definitive modality of treatment in selected cases of vascular malformations. In this article, we are presenting our experience of single stage partial/total excision of vascular malformation of various types, at different location on body without any adjuvant treatment with emphasis on the post-operative outcome. The aim of the study was to evaluate criteria for case selection, freedom from symptoms, cosmetic outcome, functional improvement, long term post-operative result and quality of life.
Methods: This study was performed to assess outcome of surgical treatment for 20 patients of vascular malformation. Fourteen were offered primary surgery and 6 out of 20 had received prior non-invasive treatment. The post-operative follow-up period was minimum 6 months and maximum for 4 years.
Results: The patients who underwent primary surgery were more satisfied than those who had received non-invasive treatment before surgery. This was because of satisfactory cosmetic outcome, minimal post-operative pain of short duration, no post-operative residual wound and minimal post-operative morbidity.
Conclusions: For treatment of vascular malformations, surgery can be offered as the primary and only treatment modality in selected cases because of immediate resolution of mass, minimal complications, satisfactory functional outcome and acceptable cosmetic appearance. Also, this improved patient compliance and long term follow up.

Keywords: Laser in vascular malformation, MRI for vascular malformation, Sclerotherapy for vascular malformation, Surgery for vascular malformation

INTRODUCTION

Vascular Malformation (VM) can be congenital or post-traumatic in etiology.[1] Congenital VM usually present in children and adolescents. These lesions commonly cause cosmetic disfigurement along with functional symptoms. The literature describes multiple pre-operative adjuvant treatment modalities. Most of these are aimed at reducing the size/extent of the lesion so as to minimize the surgical dissection during excision and reduce the chances of blood transfusion by minimizing the intra-operative bleeding. The various options are Beta blocker in children, sclerotheraphy, embolization, LASER.[2-6] The adjuvant treatment modalities need to be chosen based on age of the patient, site of the lesion, timing and duration of adjuvant treatment and finally need for surgical intervention. In this article, we have briefly touched upon the classification of the VM. This will help us in

optimizing treatment options based on their site and type of the lesion. The contrast enhanced MRI angiography of the particular region containing the lesion is very helpful in delineating the extent of the VM. MRI angiography also shows the tissue plains, vital structures in the vicinity and the organs involved. The type of the VM and the MRI angiography together guide us to select the surgical/non-surgical method of treatment.[7] Also, it helps us to identify limitations of various treatment modalities and challenges faced during the course of the treatment.[8] Here we are presenting our experience of treatment of VM with surgery as the first modality of treatment.

Table 1: Master chart.

Age (Yr) /Sex	Site	Type of VM	Previous adjuvant therapy	Surgical Excision	Complications	Follow up
6/M	Left side of face (Figure 2)	Lymphovenous	1 time STS	Debulking	Residual acceptable swelling	2year with minimal recurrence
20/ M	Left oral commissure (Figure 8)	High flow AVM	1 time STS	Primary Surgical Excision	Nil	1 year
10/ M	Right lower lip (Figure 9)	Lymphangioma	Nil	Primary Surgical Excision	Nil	1 year
40/F	Scalp	Lymhovenous	Nil	Excision & Scalp Rotation Flap	Nil	2 year
35/ M	Right buccal mucosa (Figure 10)	High flow AVM	2 time STS	Primary Surgical Excision	Nil	3 year
35/ M	Left thigh	High flow AVM	Nil	Excision & Rotation Flap	Nil	2 year
12/F	Left leg near lateral malleolus	Lymhovenous	2 STS	Primary Surgical Excision	Nil	2 year
7/M	Right thumb & radial border of forearm (Figure 4)	Lymhangiomas	Nil	Excision & Abdominal Flap	Nil	2 year
30/F	Left Forearm (Figure 5)	High flow AVM	Nil	Primary surgical excision & vein graft to radial and ulnar artery, mass of FDS of MF	Individual PIP joint flexion compensated by FDP	1 year
7/F	Right temporal extending over retro-zygomatic arch (Figure 7)	High flow AVM	Nil	Primary Surgical Excision	Right frontal nerve injury	4 year
18/ M	Right digastric triangle (Figure 14)	Lymhovenous	Nil	Excision & Rotation Flap	Postsurgical Hypertrophic scar	6 months
7/M	Left parietal scalp	High flow AVM	Nil	Excision & Rotation Flap	Nil	6 month
25/ M	Left thigh posterior aspect till deep fascia (Figure 11)	Low flow AVM	Nil	Primary Surgical Excision	Skin necrosis of 2 inch healed by secondary intention	8 months
25/ M	Right lateral quadratous femoris	Low flow AVM	3 STS	Could Not be operated due to financial constraints	-	Lost follow up after 4 months
!8/ M	Left infraspinatous (Figure 6)	Intramuscular Hemangioma	Nil	Excision of Infra-spinatous muscle	Nil	Recurrence in teres minor after 2 years with shoulder pain
14/F	Righr dorsum of foot involvin great toe (Figure 12)	Lymhangioma	Nil	Primary Surgical Excision & SSG	NIL	8 months
20/ M	Left masseter muscle & diagastric triangle (Figure 1)	High flow AVM	Nil	Primary excision of muscle & Mass extending in diagastric triangle	Initial difficulty in chewing food for a month and later recoverd	2 year
50/F	Left Cheek	Lymhovenous	1 times STS	Debulking	Residual acceptable minimal swelling	8 months
40/F	Right leg & foot (Figure 3)	Lymphovenous	Past history of debulking of leg lesion	Debulking of Leg & Foot lesion	Residual acceptable minimal swelling, one inch skin necrosis healed by secondary intention	2 years
12/F	Left index finger dorsum of PIP joint (Figure 13)	Lymphangioma	Nil	Excision & SSG	Nil	8months

METHODS

This is a short term observational study to assess outcome of surgical treatment for 19 patients operated for Vascular Malformation (VM). Total 20 patients (Table 1) were evaluated out of which 19 were operated from January 2010 to June 2018. Most of the patients were offered surgery as the primary modality of treatment although few patients (6 out of 20) had received adjuvant non-surgical treatment in the past. The patients with VM in this article span across different age groups and had VM at varying sites on the body. The minimum follow up period after surgery was 6 months and maximum of 4 years. During follow up period functional and cosmetic outcome of patient was assessed.

RESULTS

Almost 60 % of all patients with VM were aged below 20 years of age (Figure 1 and Table 1).

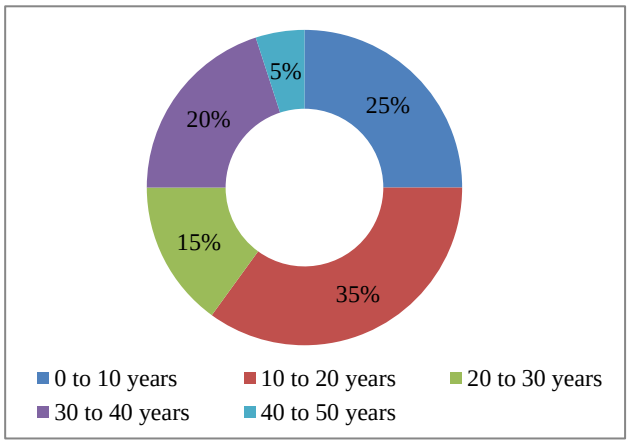

Figure 1: Age distribution.

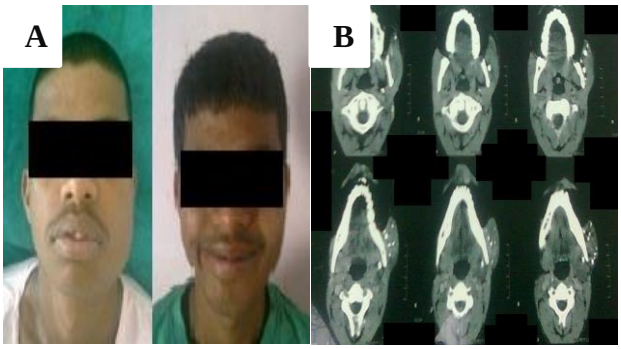

Figure 2: A & B: left cheek high flow vascular malformation involving masseter muscle (pre & post-operative image, MRI).

Out of 20 treated cases there was one case of Intra muscular Hemangioma (Figure 7) and 2 cases of low flow vascular malformation (Figure 12 and 15), 7 cases of high flow vascular malformation (Figure 2, 6, 8, 9 and 11), 4 cases of lymhangioma (Figure 5, 10 and 14) and remaining 6 cases of lympho-venous malformation (Figure 2, 4 and 13).

Figure 3: Left cheek lymphatic malformation (lympho-venous malformation).

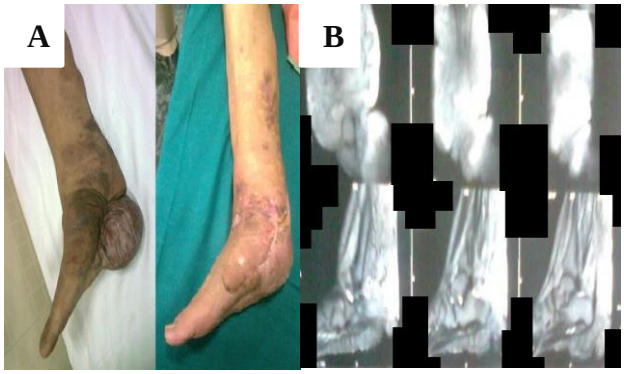

Figure 4: A & B: Right lower limb lympho-venous malformation involving foot & leg (Pre & Post-operative Image, MRI).

Figure 5: Right hand & forearm lymphangioma.

Six out of 20 patients had received adjuvant therapy in the form of 1% sodium tetra-decyl sulfate. These patients complained of pain, swelling, difficulty in movement and ulceration after sclerotherapy. Out of 6 patients 5, patients were not satisfied by the outcome of injection sclerotherapy and eventually underwent surgery. They

were satisfied by immediate result of the surgical intervention and the outcome.

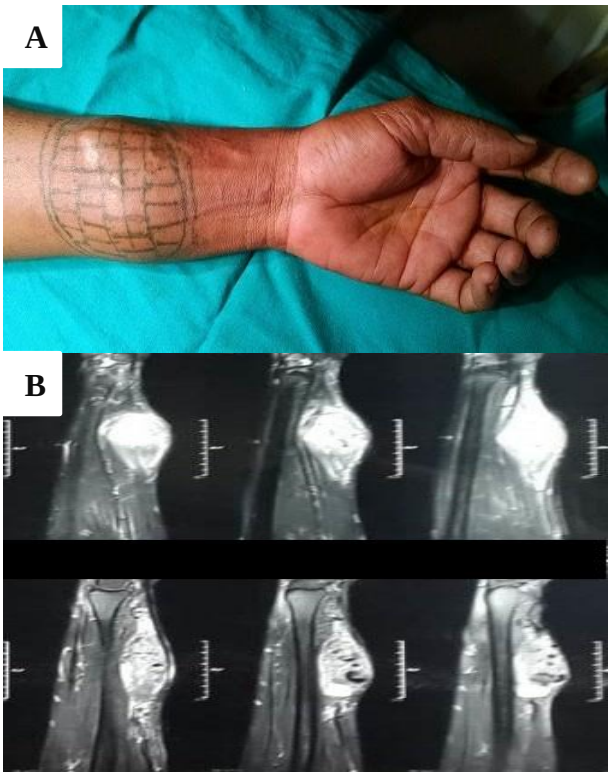

Figure 6: A & B: Left forearm high flow vascular malformation involving both radial and ulnar artery & FDS, MRI.

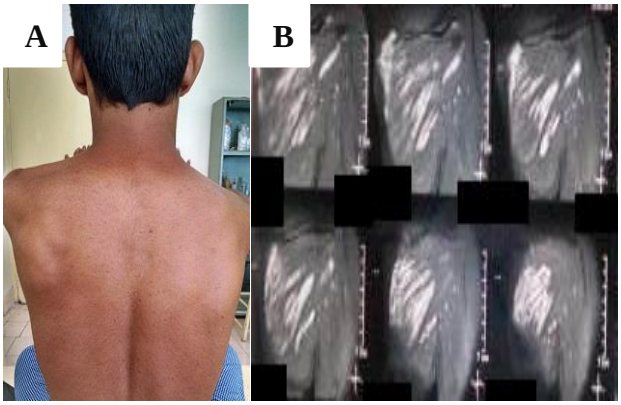

Figure 7: A & B: Hemangioma of left infra-spinatous muscle, MRI.

The patients who were offered surgery as primary mode of treatment were satisfied in view of immediate satisfactory clinical result, minimal postoperative pain, no/superficial surgical wound complications and minimal post-operative morbidity in very few cases. We offered injection scelotheraphy to 1 case of VM as patient requested for conservative mode of treatment and couldn't get operated due to financial constraints. This patient is excluded from the study.

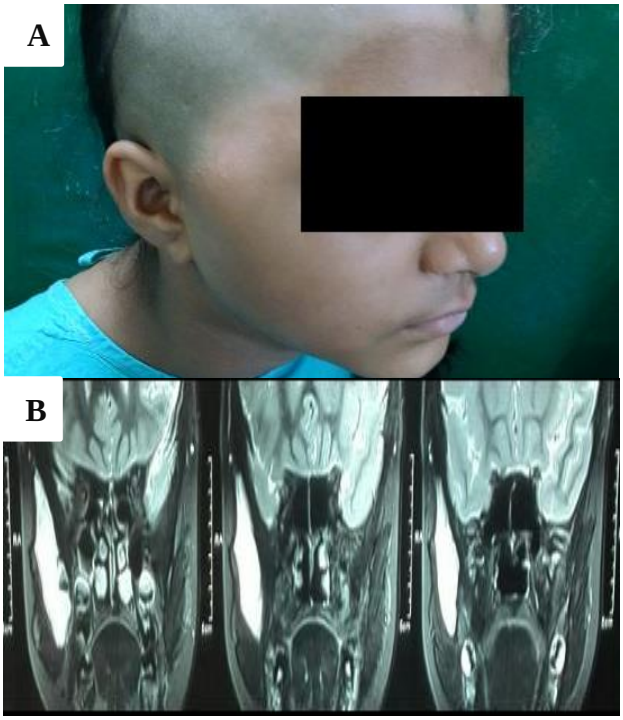

Figure 8: A & B: Right retro-zygomatic high flow vascular malformation involving pterygopalatine fossa and temporalis region in a 7-year-old female child, MRI.

Figure 9: Left side oral commissure High flow VM.

Figure 10: Lymphangioma on right lower lip near oral commissure.

Figure 11: Right buccal region intra-oral high flow vascular malformation.

Figure 12: Left thigh low flow vascular malformation.

Figure 13: Lymphatic malformation on dorsum of left foot.

Figure 14: Lymphangioma on left index finger.

Figure 15: Low flow VM in right diagastric triangle.

DISCUSSION

Vascular tumors are mainly classified into hemangiomas and vascular malformations. Haemangiomas are characterized by a history of rapid neonatal growth, endothelial hyperplasia during the proliferative phase and fibrosis with reduced cellularity during the involutionary phase. On the other hand, vascular malformations are characterized by being present at birth, growing with the child, and have a normal rate of endothelial turnover.[9,10]

The patient with vascular malformation usually present with pain, functional impairment, aesthetic disfigurement of the affected body part and rarely, bleeding.[11]

There is no fixed documented and easy road map during the selection of various treatment modaliies for vascular malformation of any sites in body.[12,13] The basic factors which decide the treatment modality in vascular tumors/ malformations are:

- Age of the patient
- Site of the lesion

- Involvement of the surrounding vital structure/s with documentation on contrast enhanced MRI scan
- Surgeons past experience of treating VM
- Patient's and their relatives' knowledge about the disease/ syndrome and willingness for treatment and long term follow up.

The common non-surgical methods for treatment of various vascular malformations are LASER (Nd:YAG) and 5% monoethanolamine oleate sclerotherapy.[3,4] There are advantages and disadvantages for both surgical and nonsurgical methods of treatment of large vascular malformations. The main advantage of LASER is in cutaneous vascular malformations like port wine stain and vascular malformations in paediatric age groups.[8,14] The disadvantages of the laser photocoagulation are multiple sitting therapy, blistering, tissue necrosis leading to bleeding, tissue edema, intraoral infection, hyper/hypo-pigmentation.[15] The main reported complications of sclerotheraphy are renal, nerve and muscle damage along with pulmonary embolism and cardiovascular collapse.[16]

The main disadvantages of surgical therapy are intra-operative bleeding, injury to nerve/ surrounding structure and post-operative scar.

The patient selection for surgery is based on the site and size of mass in tissue and age of the patient. The surgical approach to any vascular malformation has to be individualized based on clinical examination of the patient, radiological images, intra-operative findings and review of literature on results of surgical treatment of VM. The primary surgical excision of the VM is done in view of the cost factor and non availability of the treatment by interventional angiography and embolization for preoperative reduction in the size of the lesion.[17]

The main reason of preferring the adjuvant therapy before surgical intervention was to reduce the size of the swelling by minimizing its blood supply and directly reduce the amount of blood loss during surgery. In children, the high flow vascular malformation with hypertension and those with ulceration and bleeding are treated initially by B- blocker and embolization. During surgery, the blood loss from VM distal to single bone on the extremity can be controlled by pressure guided tourniquet. There are other measures of minimizing the blood loss like intra-operative hypotension, bipolar electro-cautery, good surgical expertise and knowledge of Anatomy of the feeder vessel supplying the organ affected by vascular tumor. The main aim of this article is to present our experience of 20 various cases of vascular malformations affected at different parts of the body, in various age group and criteria for selection for the primary surgical intervention without any adjuvant therapy.[17,18]

The parents of the patients in pediatric age group are reluctant to prefer the surgical mode of treatment and most of the time the parents make enquiry about the non-surgical adjuvant therapy. Till the parents/guardian reach the decision about the surgical mode of treatment, they approach multiple consultants for opinion. It is also noticed that those patients who took time to make decision about any mode of treatment and approached multiple consultant, the size of VM had increased on contrast MRI scan film with involvement of surrounding structures. The same is observed in this study as 5 pediatric and 1 adult patients received adjuvant treatment.

CONCLUSION

During this study, it was observed that patients preferred primary surgery over adjuvant treatment for the treatment of VM because of immediate resolution of mass, lesser complications, good functional outcome and acceptable cosmetic appearance. This was also helpful for long term patient compliance and follow up. Therefore, the patients with VM may be offered surgery as the primary treatment modality based on following criteria for case selection:

- Well localized tumor mass,
- Vascular anatomy well established by MR angiography,
- Feasibility of control/ligation of the feeder vessel during surgery,
- Well maintained surgical plains available between the structures involved by the VM and surrounding normal unaffected structures.
- Possibility of minimal blood loss by use of intra-operative hypotension, tourniquet and bipolar electro-cautery.

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