Research Article

Vascular Malformations of Head and Neck Region: Our Experience at Mayo Hospital, Lahore.

Afzaal Bashir¹, Muhammad Mustehsan Bashir², Muhammad Sohail³

¹Assistant Professor, Department of Plastic and Reconstructive Surgery, KEMU/ Mayo Hospital, Lahore; ²Chairman, Department of Plastic and Reconstructive Surgery, KEMU/ Mayo Hospital, Lahore; ³Associate Professor, Department of Plastic and Reconstructive Surgery, KEMU/ Mayo Hospital, Lahore.

Abstract

Background: Vascular malformations of head and neck region are present in approximately 0.3% to 0.5% population and usually noticed in early childhood. They keep on growing in proportion to growth of the child with no regression. For management point of view, vascular malformations are categorized into low flow (Lymphatic, Capillary and Venus) and high flow. Diagnosis of vascular malformation is established by clinical examination easily but sometimes, regardless of clinical signs, radiological adjuncts (Doppler, MRI) are mandatory. Treatment option depends upon size and type of malformation (low or high flow).

Objective: To study different types of vascular malformations in head and neck region and their response to various treatments in our population.

Methodology: This prospective study was conducted in Plastic Surgery Department, Mayo Hospital, Lahore, Pakistan, from Feb 2017 to Jan 2018. Doppler was done in all patients; MRI was done where necessary to find out extent of the malformation while CT angiography in few cases to map out feeding vessels. Treatment was based on type, size and location of the malformation. Compression, sclerotherapy, LASER and surgical de-bulking alone or in combination were among the treatments.

Results: A total of 140 patients were included out of which 79(56%) were males and 61(44%) were females. Mean age was $22(\pm 16)$ years (range 6-50 years). Low flow malformations were 98(70%) out of which 27(19%) were capillary, 31(22%) venous and 40(28%) were combined. High flow malformations were in 42(30%) where 9(6%) were arterial and 33(25%) arteriovenous. Operative treatment and adjuncts, alone or in combination, were employed depending on type of malformation. Satisfaction with outcome was more in patients having combination therapy. Non-surgical modalities, when used alone, were not sufficient to produce desired results

Conclusion: Surgical debulking, with or without non surgical technique, proved to be the mainstay to have good outcome.

Corresponding Author | Dr. Afzaal Bashir, Assistant Professor, Department of Plastic and Reconstructive Surgery, KEMU/ Mayo Hospital, Lahore **Email:** dr.afzaal@yahoo.com

Kev Words: Vascular malformation, Sclerotherapy, LASER, debulking, high flow, low flow

Introduction

Vascular anomalies have been very aptly classified by Mulliken and Glowacki into

vascular tumors and vascular malformation to abate the confusion in nomenclature. Vascular tumors include infantile haemangioma, congenital haemangioma, tufted angioma, hemangiopericytoma, pyogenic granuloma, kaposiform hemangioendothelioma etc. Vascular malformations are present in approximately 0.3% to 0.5% population and their presence is usually noticed in early childhood as a small compressible soft tissue swelling which keeps on growing in proportion to growth of the child with no regression.¹

Comprising of abnormally formed dilated vascular channels lined by quiescent epithelium, vascular malformations can be categorized radiologically into low flow (Lymphatic, Capillary and Venus) and high flow (Arterial) malformations. Combinations of these varieties in various proportions may also exist.² Behavior, progression, management options and final outcome depends on category of the lesion. That is why, for counseling as well as planning any treatment, we need to establish exact diagnosis of type of malformation.³

Single or multiple sites on body ranging from vertex to toe can be involved but in head and neck region, vascular malformations are very common. Where they culminate into significant disfigurement and large number of complications are associated with their natural course like recurrent bleeding, thrombosis, ulceration and infection. These complications usually occur quite late in the course of these lesions. Repeated hospital visits and admissions are needed which are not only cumbersome for the patient but also overburden the surgical team.⁴

Diagnosis of vascular malformation can be established by clinical examination easily but sometimes, regardless of clinical signs, radiological adjuncts (Doppler, MRI) are mandatory to confirm the diagnosis. These radiological investigations also help in clarifying depth and relations of the lesion with its surrounding structures.⁴

Common complications associated with the disease are pain, ulceration, bleeding and cosmetic disfigurement. Treatment option for vascular malformations depends upon size and type of malformation (low or high flow), associated complications, age of the patient and stage of presentation.^{5,6} Treatment options include surgical debulking, injection sclerotherapy and LASER.

These options can be employed alone or in combination. Despite of all the stratification and individualized modern care, wrong diagnosis, diagnostic delay, high recurrence and complication rate have been noticed particularly in head and neck region. So far, only few studies available addressing vascular malformations of head and neck region. This prospective study was conducted in our unit to have a better understanding of characteristics of vascular malformation, age of presentation, importance of radiological investigations, treatment plans, post-operative complications and recurrence rate in this region. See the strategies of the s

Methods

In Plastic Surgery Department, Mayo Hospital, Lahore, Pakistan, this prospective study was conducted from Feb 2017 to Jan 2018 with a sample size calculated as 140 cases of vascular malformations of head and neck region diagnosed and picked up from out-patient department. Patients with diagnosis of hemangioma or other vascular tumors as hemangioendothelioma and angiosarcoma were excluded.

Demographic data like age, sex of patient, presenting complaints like aesthetic as well as functional derangement, pre procedure complications and characteristics of vascular malformation like sub-area of head and neck involved, type of vascular malformation and past treatment taken were noted. After counseling and informed consent, doppler study was done in all patients to distingueish high flow from low flow malformations. MRI was also done in patients where it was necessary to find out extent of the malformation, involvement of neighboring structures and treatment planning particularly tongue and floor of mouth. In cases having high flow malformations, CT angiography was performed to map out feeding vessels. Conventional angiography accompanyied by Super-selective gel foam embolization was done in high flow malformation to minimize blood loss during the follow up surgery after 36 to 48 hours.

Treatment was based on type, size and location of the malformation. Lesions with smaller size allowing primary closure were excise. Arterial malformations were excised with affected overlying skin, venous malformations were debulked with combination of injection sclerotherapy. LASER was offered for capillary malformations. Sclerotherapy using 3% Sodium Tetradecyl ulphate injections was done for the patients with venous malformations in a dose of 0.5ml/kg and was followed by compression garment in the form of head band, chin belt and cheek support garment. Injection was repeated after four weeks interval if required and was followed by surgery if needed. Cases with capillary malformations benefited from LASER treatment which was done in 4 to 9 visits with 4 to 6 week interval. All surgeries were performed under general anesthesia. Co-amoxiclave 1G in single dose was given intravenously to all patients at the time of induction. After marking the lesions, adrenaline in the concentration of 1:100,000 was infiltrated. Multi-planner Debulking was done haemostasis secured, wounds closed, compression dressing was done and all specimens were submitted for histological evidence of diagnosis. Patients got discharged on 6th to 10th day and were put on regular follow up.

Complications associated with sclerotherapy were mainly pain (scored by visual analogue scale), ulceration (defined as break in epidermis of overlying skin) and necrosis of skin (defined as destruction of skin due to hampered blood supply). Complications noted in association with de-bulking were bleeding (defined as oozing of blood which required wound re-exploring and dealing with the bleeding vessel), hematoma (defined as blood collection leading to pain and swelling), infection (defined as redness and purulent discharge from suture line along with temperature more than 100° F), wound dehiscence (defined as complete or partial opening of sutured wound) and recurrence. Laser therapy was having concerns of repeated sessions.

After getting IRB/Ethical approval vide letter number 1997/RC/KEMU, information was recorded using a proforma. Quantitative variables like age and pain score were projected in the form of mean (SD) while categorical variables including gender, type of malformation and complications were shown as percentages and frequencies. All the data was analyzed by SPSS 21 and p- Value <0.05 was taken as significant.

Results

A total of 140 patients were included out of which 79(56%) were males and 61(44%). Mean age was 22(16) years. Referral from other departments was in 40(28%) patients out which 11(8%) were with wrong diagnosis. Low flow malformations were 98(70%) out of which 27(19%) were capillary, 31(22%) venous and 40(28%) were combined. There was no purely lymphatic malformation seen. High flow malformations were in 42(30%) where 9(6%) were arterial and 33(25%) arteriovenous.

Tongue and floor of mouth was involved in 28(20%), cheeks in 19(13%), lips in 57(40%), scalp in 22(16 %) and involvement of neck was noticed in 14(10 %) patients.

Cosmetic disfigurement was the main concern in all patients (100%) with face and neck involvement while drooling of saliva and difficulty in speaking and eating was the chief complaint in patients with lip, tongue and floor of mouth involvement.

Color Doppler was performed in all (100%) patients, Magnetic Resonance Imaging (MRI) was done in 43%, CT Angiography was performed in 19%, Conventional Angiography with Gel Foam Embolization was done in 3% patients to get forehand control which helped in minimizing bleeding during surgery planned within coming 36 to 48 hours.⁸

Surgery alone in the form of debulking was performed in 47% patients while in 26% patients it was done in combination with sclerotherapy before or after the surgery. Sclerotherapy alone was administered in 17% patients having venous malformation. Most of the patients with combination therapy were satisfied with final outcome as compared to the patients dealt with surgery alone. Significance was shown by independent t-test with p value less than 0.05.

LASER benefited in 13% patients who were having capillary malformation. Post operative mean pain score was 7(2.10). In operated cases, oedema was found in all (100) patients, wound dehiscence in 5%, infection in 2% and bleeding in

4%, haematoma in 2%, skin necrosis in 4%, scar widening in 7% and recurrence in 9% patients.

After sclerotherapy, marked oedema was noticed in all (100%) patients in first 24 hours which progressively settled in two to three weeks time. Ulceration occurred in 13%, bleeding in 6%, skin necrosis in 10%, infection in 2% and recurrence in 18% patients.

LASER burns in 2% and spot pigmentary change noted in 5% patient dealt with LASER therapy.

Marked Cosmetic as well as functional improvement was noted in all cases. Satisfaction level of patients was very high who were treated surgically while recurrence was 18% patients after sclerotherapy alone who later required surgery and 7% cases handled with surgery alone needed revision at or after 6 months. All the patients were kept in regular follow up and no patient was dropped out.



Figure 1: Pre and Post operative pictures of Arterial malformation
(A-Pre-operative Arterial Malformation Lt.

Forehead, B-Post-operative Arterial Malformation Lt. Forehead, C-Pre-operative Arterial Malformation Rt.Cheek, D-Post-operative Arterial Malformation Rt.Cheek)



Figure 2: Pre and Post Operative Pictures of Venous Malformation Lip (A-Pre-operative Venous Malformation Upper Lip, B-Post-operative Venous Malformation Upper Lip, C-Pre-operative Venous

Malformation Lower Lip, D-Post-operative Venous Malformation Lower Lip)



Figure 3: Pre-operative (A) and Post Operative (B) picture of combined Low flow Malformation Forehead



Figure 4: Pre-Sclerotherapy (A) and Post-Sclerotherapy (B) picture of Venous Malformation Cheek





Figure5: Wound Dehiscence (A) and its Outcome (B) on Conservative Management

Discussion:

Most of the malformations (83%) were noticed at birth while some (17%) were recognized later.¹¹ However 14 years was the mean age of seeking medical help which is most likely due to unawareness and lack of facilities at periphery.¹²

Studies represent low flow malformations to be the most prevalent.⁴ In our patients, low flow malformation was (79%) compared to high flow malformation (21%) and out of these Low flow, combined malformations predominated (56%) followed by venous malformations alone (21%) and then alone capillary (14%). In contrast to literature, lymphatic formations were noticed in combined form only and not found purely even in a single case.¹³

Similar to published literature, radiologic investigations (Color doppler, MRI, CT Scan) were very useful for confirmation of the diagnosis, classification and finding relations of malformations with surrounding structures which ultimately helped in planning the treatment.¹

According to the published data, choice of treatment (Non-operative, Operative or combination) depends upon type of malformation, extent, depth, flow pattern and size of lesion. Combination of operative treatment and adjuncts (Sclerotherapy, LASER, Compression, Embolization) is usually needed. Sclerotherapy, done by 3% STD, is the best choice for venous malformations while capillary malformations require LASER for skin color improvement. 4,6,9,10

In this study we also found out that non-surgical modalities, when used alone, are not sufficient to produce desired results rather they are employed with surgery to give better outcome leading to more satisfaction of patient with final outcome. Surgical debulking is the most important and the valuable gadget in the armamentarium. Small lesions can even be excised completely and wounds are closed primarily while lager lesions may require multiple debulkings and sometimes coverage in the form of skin graft or flap is needed to close the wound. In our study, no patient needed flap coverage and one patient required skin graft on forehead. Per-operative blood loss can be minimized by combining sclerotherapy in low flow and superselective embolization in high flow malform-ations. 12

Post-surgical complications noticed were comparable to the published stuff. Oedema was found in 100% patients which resolved in one week time spontaneously. Surgery of high flow malformations led to bleeding in 4% patients in first 48 hours as shown by other studies. Similar to other studies, wound infection and dehiscence settled conservatively and wound infections was noted in 5% patients leading to wound dehiscence and required antibiotics. ^{7,9,10}

Patients not complying with scar management therapy, 7% presented with scar widening and hypertrophy. Most of the patients (67%) were having satisfactory results. Recurrence is reported to be common with low flow malformations and in our study occurred in 9% cases and all of them were with low flow malformations.⁶ Our study was having limitations regarding no management of lymphatic malformations and no patient turned out to be with having this issue purely.

Conclusion:

The conclusion of our study was that surgical debulking with or without non surgical technique proved to be the mainstay in management of high as well as low flow malformations of head and neck region while laser was found suitable for capillary malformations.

Ethical Approval: Given

Conflict of Interest: The authors declare no conflict of interest

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References:

- 1. Beckers K, Faes J, Deprest J, Delaere PR, Hens G, De Catte L, et al. Long-term outcome of pre-and perinatal management of congenital head and neck tumors and malformations. International journal of pediatric otorhinolaryngology. 2019;121(1):164-172.
- 2. Park H, Kim JS, Park H, Kim JY, Huh S, Lee JM, et al. Venous malformations of the head and neck: A retrospective review of 82 cases. Archives of plastic surgery. 2019;46(1):23.
- 3. Prakash S, Das S, Suri R. Unilateral facial arteriovenous communication and its clinical implications. European Journal of Anatomy. 2019;9(2):107-109.
- 4. Vaughn J, editor Emergency Imaging of the Nontraumatic Pediatric Head and Neck. Seminars in Ultrasound, CT and MRI. Elsevier. 2019.
- 5. Shwayder T, Schneider SL, Icecreamwala D, Jahnke MN. Capillary Malformations. Longitudinal Observation of Pediatric Dermatology Patients. Springer. 2019.
- 6. Prasad R, Kumar A, Kumar A, Kumar V. Observation on vascular malformation in respect of sclerotherapy as a treatment of choice. Head and neck. 2019;3(1):8-13.

- 7. Nilesh K, Reddy S, Tewary S, Zope SA, Vande AV. Sclerotherapy-A conservative approach for the management of compound hemangioma of the upper lip in a young female patient. International Journal of Contemporary Dentistry. 2019; 9(1):1-5.
- 8. Lin M-y, Lin C-s, Hu S, Chang J-m, Chung W-h, Zhang Z-y, et al. The application of 595-nm pulsed dye laser for vascular anomalies in a Chinese population: a 10-year experience. Journal of Cosmetic and Laser Therapy. 2019; 21(3):171-178.
- 9. Saeed W, Bashir B, Altaf F. Late onset arteriovenous malformation of nose: a clinical report. Journal of Pakistan Association of Dermatology. 2019;29 (2):267-269.
- 10. Kuriakose MA, Chakrabarti S, Cheong SC, Kowalski LP, Novaes Pinheiro T, Farah CS. Head and Neck Tumors. Contemporary Oral Medicine: A Comprehensive Approach to Clinical Practice. Springer; 2019.
- 11. Hunasgi S, Koneru A, Vanishree M, Manvikar V. Giant arteriovenous malformation of the face and upper lip. Journal of Oral and Maxillofacial Pathology. 2019;23(4):32.
- 12. Flors L, Hagspiel K, Park A, Norton P, Leiva-Salinas C. Vascular malformations and tumors. Part 2: Low-flow lesions. Radiología (English Edition). 2019;61(2):124-33.
- 13. Bourne Jr MH, Kern RM, White D, Balak-rishnan K. Tracheal Submucosal Lymphovenous Malformation. American journal of respiratory and critical care medicine. 2019;199(4):26-27.