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A STUDY OF VASCULAR ANOMALIES AND THEIR MANAGEMENT

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ABSTRACT

Background: A clinical study of vascular anomalies and their management in a tertiary care center in Telangana. Materials and Methods: Prospective and retrospective study in patients admitted in the Department of Plastic and Reconstructive Surgery with vascular anomalies within the stipulated time period of 48 months. **Result:** The Age distribution were 1-10 years is 28.0%, 11-20 years is 12.0%, 21-30 years is 48.0%, >30 years is 12.0%. As per the stats, males were affected more than the females (60% > 40%). The number of lesions distribution were 1 is 96.0%, 3 is 4.0%. The Outcome distribution were Upper lip has highest percentage (12.0%), and all other variables except Right cheek has lowest percentage (4.0%). The Course distribution were I & P is 88.0%, I & P following trauma is 4.0%, Sudden onset is 8.0%. The Complications distribution were No has highest percentage (80.0%), and all other variables has lowest percentage (4.0%). Out of the 25 cases that presented to our OPD, none of them were hemangioma, 5 cases of venous malformations were identified, 1 case of lymphatico-venous malformation and 19 cases of arterio-venous malformations were diagnosed. The Treatment distribution were Excision has highest percentage (12.0%) and all other variables except Sclerotherapy has lowest percentage (4.0%). The Surgery distribution were No is 12.0%, Previously done in childhood is 4.0%, Yes is 84.0%. The Complication distribution were No has highest percentage (68.0%) and all other variables except recurrence has lowest percentage (4.0%). The recurrence distribution were No is 76.0%, Yes is 24.0%. Conclusion: Sclerotherapy was effective for lesions of dimensions less than 2x2cm and no recurrence pattern was noted in those cases in the follow up duration of 1 year. Whereas sclerotherapy followed by excision was mostly done for diffuse lesions and hence recurrence was noted in all the cases.

INTRODUCTION

Vascular anomalies include a spectrum of disorders including vascular tumors and vascular malformations. Vascular anomalies were first appropriately classified by Mulliken and Glowacki in 1982 which was based on the biological activity of the tumor and helped in understanding and classifying vascular malformations. Prior to that hemangiomas were often called cherry angioma, strawberry angioma or cavernous angioma. Molecular studies suggest that vascular anomalies are caused by dysfunctional signaling processes that regulate proliferation, apoptosis, differentiation, maturation and adhesion of vascular cells.^[1]

A hemangioma is an abnormal proliferation of blood vessels occurring in any vascularized tissue. There

seems to be consensus that the term "hemangioma" should refer to "hemangiomas of infancy," which have a predictable natural history that includes absence at birth followed by a period of growth over 6-18 months and then a period of involution that may take several years. Hemangiomas are the most common cutaneous tumor of infancy and demonstrate rapid growth followed by a low spontaneous involution or resolution in 5-7 years. Vascular malformations on the other hand enlarge with the growth of the child and do not undergo spontaneous involution. Vascular malformations are caused by the disturbance in the late stages of angiogenesis and result in persistence of arteriovenous anastomosis present during embryonic life. They may be capillary, lymphatic, venous, arterial or mixed.^[2]

Vascular malformations have been recognized throughout history as birthmarks. In 1866, Dugas conducted a scientific analysis of birthmarks and concluded that they were caused by defects in embryonic development. Virchow and Wagner,^[3] established the first classification system of vascular malformations, based on channel architecture and histomorphologic appearance.

MATERIALS AND METHODS

Clinical study in patients admitted in the Department of Plastic and Reconstructive Surgery with vascular anomalies within the stipulated time period of 48 months (2 years retrospective and 2 years prospective from February 2021 to May 2024) are included in the study. 48 months (2 years retrospective and 2 years prospective).

All patients with vascular anomalies admitted in the Department of Plastic and Reconstructive Surgery in Osmania General Hospital, Hyderabad within the stipulated time period is included in the study. Retrospective analysis includes old case records and photos.

Inclusion Criteria

All patients with vascular anomalies admitted in the Department of Plastic and Reconstructive Surgery willing to participate in the study are included.

Exclusion Criteria

Patients who are unfit for anesthesia and with CNS AV Malformations.

The collected data were entered in the Microsoft Excel 2016 and analysed with IBM SPSS Statistics for Windows, Version 29.0. (Armonk, NY: IBM Corp). To describe about the data descriptive statistics frequency analysis, percentage analysis were used for categorical variables and the mean & S.D were used for continuous variables.

RESULTS

The [Table 1] shows Age distribution were 1-10 years is 28.0%, 11-20 years is 12.0%, 21-30 years is 48.0%, >30 years is 12.0%. Gender distribution were Female is 40.0%, Male is 60.0%. Number of Lesions distribution were 1 is 96.0%, 3 is 4.0%.

Table 1: Demographic distribution			
Age distribution	Frequency	Percent	
1 - 10 yrs	7	.28.0	
11 - 20 yrs	3	.12.0	
21 - 30 yrs	12	48.0	
Above 30 yrs	3	12.0	
Total	25	100.0	
Gender			
Female	10	40.0	
Male	15	60.0	
Number of Lesions distribution			
1.0	24	96.0	
3.0	1	4.0	

Table 2: Distribution of the Lesions and course.

Distribution of the lesions	Frequency	Percent
Chin	1	4.0
Hard palate	1	4.0
L forearm	1	.4.0
L little finger	1	.4.0
L Loin	1	4.0
L nasal	1	4.0
L thumb	1	4.0
L upper eyelid	1	4.0
Nose	1	4.0
Philtrum and upper lip	1	.4.0
R cheek	2	.8.0
R forearm	1	4.0
R Gastrocnemius	1	4.0
R lower lip, R cheek, Tongue Pharynx	.1	4.0
R preauricular	1	4.0
R scalp	1	4.0
R temporal	1	4.0
R thumb	1	4.0
R upper lip	1	4.0
Soft	1	4.0
tongue	1	4.0
Upper lip	3	12.0
Total	25	100.0
Location		
Head & Neck	18	72%
Upper Limb	5	20%

Trunk	1	4%
Lower Limb	1	4%
Course distribution		
Insidious in onset	21	84.0
Following trauma	2	8.0
Sudden onset	2	8.0

Distribution of the lesions and Upper lip has highest percentage (12.0%), and all other variables except R cheek has lowest percentage (4.0%). Distribution was I & P is 84.0%, I & P following trauma is 8.0%, Sudden onset is 8.0%.

Table 3: Complications distribution			
Complications	Frequency	Percent	
No	20	80.0	
Bleeding	1	4.0	
Difficulty in speech and difficulty in closing mouth	1	4.0	
Difficulty in deglutition	1	4.0	
Recurrence	1	4.0	
Recurrence history post excision 2 yrs ago	1	4.0	
Total	25	100.0	

The above table shows Complications distribution were No has highest percentage (80.0%), and all other variables has lowest percentage (4.0%).

Treatment distribution	Frequency	Percent
Sclerotherapy alone	5	20.0
Sclerotherapy followed by excision	5	-20.0
Excision alone	14	56.0
Excision with flap cover	1	4.0
Total	25	100.0
Surgery distribution		
No	3	12.0
Previously done in childhood	1	4.0
"Yes	21	84.0

The above table shows Treatment distribution were Excision has highest percentage (12.0%) and all other variables except Sclerotherapy has lowest percentage (4.0%). Surgery distribution were No is 12.0%, Previously done in childhood is 4.0%, Yes is 84.0%.

Table 5: Complication and resurrence distribution			
Complication	Frequency	Percent	
Flap Necrosis Bleeding Recurrance	1	4.0	
L little finger extension weakness	1	4.0	
Recurrance	5	20.0	
Recurrance Flap necrosis	1	4.0	
No	17	68.0	
Recurrence distribution			
No	19	76.0	
Yes	6	24.0	

The above table shows Complication distribution were No has highest percentage (68.0%) and all other variables except Recurrence has lowest percentage (4.0%). Recurrance distribution were No is 76.0%, Yes is 24.0%.

Table 6: Descriptive Statistics				
Descriptive Statistics	.Minimum	Maximum	Mean	.SD
Age	1.0	59.0	21.1	14.0
Age Of Onset	08	49.00	16.9	13.3
Duration	.66	26.00	4.4	5.2
Number of Lesions	1.0	3.0	1.1	0.4
Lesion Type And Size	.1.0	20.0	6.1	5.1
CBP	8.9	14.1	11.5	1.4

The above table shows Descriptive Statistics of Age, Age of Onset, Duration, number of Lesions, Lesion Type and Size, CBP.

Table 7: Type of Lesion Distribution			
Type of Lesion	Frequency	Percent	
Venous Malformation	5	-20	
Lymphatico-venous	1	4	
Arterio-venous	19	.76	
Total	25	100	

INTRA OP



Post Op sclerotherapy (1 sitting) followed by excision of Vascular Malformation of upper lip- Vessel: Supeior Labial Artery branch

DISCUSSION

In our study a total of 25 cases were observed all of which presented with vascular malformations and none with hemangioma, the confirmation of which was done by CT/MR Angiography and post excision biopsy. Vascular malformations though present since birth, presenting age was only after 10 years of age as per previous studies, but in our study majority of the cases presented to the OPD with complaints in the age group 21-30 years. Sachin et al. reported that haemangiomas were only 15.56% of the vascular anomalies in their study,^[4] and 31.59% in a study by Ye et al.^[5] The male-to-female ratio among

haemangiomas in our study was 1:2.16 which is less than what is seen in European population.^[6] Lesser female predominance of 1:1.49 was also reported by Ye et al.^[5] Regarding regional predilection, majority (60.2%) of the lesions in the present study were cervico-facial followed by extremities and trunk which is reflected in other studies as well.^[4,5]

The reason behind the late presentation could be ignorance or difficulty to come to the hospital as we are mostly dealing with majority of the lower middle class to lower class population and also because sometimes the lesions become evident after a traumatic incident which in our study happened with 2 of the cases- Vascular malformation of upper lip and vascular malformation involving the right gastrocnemius muscle which presented with achilles tendon shortening and muscle wasting.

Vascular malformations are localized or diffuse errors of embryonic development of which port-wine stain is a common vascular anomaly that is present at birth and persists throughout life. In our study head and neck was the most common site involved in 18 cases (72%), of which upper lip involvement was the most common site which included 3 cases (12%), followed by extremity involvement which is upper limb in 5 cases (20%) while 1 (4%) had lower limb involvement and 1 (4%) had trunk and loin involvement. Of the cheek involvement in 3 cases, extensive and bilateral involvement was present in 1 case, the rest 2 cases had unilateral involvement. The adjacent mucosal involvement was present in 2 cases out of the 3 cases with cheek involvement.

In our study of the 18 cases with head and neck involvement involvement none of the cases had ocular involvement or CNS involvement or both. The case with extensive involvement of R lower lip, R cheek, tongue and pharynx, external carotid artery was found to be the vascular supply to the lesion but CT brain findings were found to be normal. In the case of Vascular malformation involving the scalp, the CT Brain findings were normal and the involved vessel supplying the lesion was found to be superficial temporal artery excision of which was done and recurrance was identified 1 year post excision. In our study none of the cases presented with any syndromic association nor did they have any positive family history of vascular lesions. Even as we know that vascular malformations are congenital conditions which becomes evident or are noticed after any traumatic incident, the study has clarified the same with majority of the patients presenting with the complaints in the age group 21-30 years. The major complications associated with these conditions with which the patient presented to our OPD included mostly cosmetic issues and in 2 of the cases (vascular

malformation of soft palate and extensive vascular malformation of right cheek with pharynx and external carotid artery as feeder vessel) presented with difficulty in deglutition and bleeding episodes.

Among the 25 cases only surgical debulking or excision was done for 15 cases (60%) and all these lesions were either more than 2x2cm in dimensions or the patient requested for only surgery and no sclerotherapy trial. Among the 25 cases only sclerotherapy was done for either lesions of dimensions less than 2x2cm which were in total 3 cases and 1 inoperable case and 1 case where the patient requested for only sclerotherapy (vascular malformation of philtrum and upper lip of dimension 4x2cm)

Sclerotherapy alone was done for 3 of the cases and recurrence were not observed in them during the follow up period of 1 year. Sclerotherapy multiple sittings followed by excision was done for 5 cases of which recurrance was noted for all the cases. Recurrance was noted in 7 of the operated cases and flap necrosis was noted in 2 of the cases which included vascular malformation of the thumb and palate, extension weakness was noted post op in case of vascular malformation of left forearm which improved with regular physiotherapy. Several sclerotherapy options are available for sclerotherapy of LMs, namely, ethanol, bleomycin, OK-432 and doxycycline.^[7-9] In our series, we have used bleomycin only. There was no sclerosant-related complications in LMs. Carbon dioxide laser has also been reported to be used in intra-oral LMs.^[10] The management of VMs is also a multimodality approach which includes surgery, neodymium-doped yttrium aluminium garnet (Nd:Yag) laser and sclerotherapy.^[11] Nd:Yag laser is reported to be very effective as it causes shrinkage and thrombosis of the aberrant venous channels.^[11] Sclerotherapy is also an effective modality for VMs, and the sclerosants commonly used are sodium tetradecyl sulphate, ethanol, bleomycin and OK-432.^[12,13] In our series, complete ablation of most of the VMs was possible with surgical excision either primarily or after sclerotherapy. However, for VMs in superficial plane, especially in face and hand, only sclerotherapy was tried at first to avoid post-surgical scarring and deformity. The gold standard of management of highflow or AVM is excision with or without preoperative embolisation of feeding vessel.^[14]

CONCLUSION

Sclerotherapy was effective for lesions of dimensions less than 2x2cm and no recurrence pattern was noted in those cases in the follow up duration of 1 year. Whereas sclerotherapy followed by excision was mostly done for diffuse lesions and hence recurrence was noted in all the cases.

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