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Role of Surgical Intervention either Excision or Injection of Sclerotherapy in Treatment of Infantile Haemangioma

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ABSTRACT

Background: Infantile Haemangioma (IH) is a benign vascular tumor characterized by endothelial cell proliferation of blood vessels in skin or visceral organs and generally lesion enlarges in what is called proliferative phase and overall growth by 9 months age. The present study aimed to detect the best time of surgical intervention in management of infantile hemangioma.

Patients and methods: A prospective clinical trial study was included in carried out in Zagazig University Hospitals. Patients with infantile haemangioma full filing inclusion criteria were evaluated and received medical treatment with beta blockers if not complicated and followed up for possibility of surgical intervention.

Results: Major site was forehead with 25% followed by back & scalp each with16.7% then abdomen, abdomen & back and Lip with 8.3% each and finally arm, buttocks, chest wall and face with percentage of 4.2%. The majority were good with 37.5%, excellent with 29.2% and bad with 16.7%. Bleeding reported at 4 cases with 16.7% ulceration and infection 8.3%. Medical management was in 66.7% and surgical in 33.3% (8 cases), 6 cases were excision and 2 cases had injection (one of them need excision) and combined were in 29.2% . There was significant decrease at all time from pre and 2 month and from pre to 4 month and from pre to 6 month. There were significantly associated with surgical intervention also irregular shape, firm consistency and multiple lesion were significantly associated with surgical method. Surgical management significantly associated with better outcome.

Conclusion: Surgery procedure for IH may be considered when there are likely to be significant functional or cosmetic deficits despite medical therapy or when these deficits might be more severe with the prolonged treatment time frame associated with medical options.

Keywords: Infantile Haemangioma; Excision; Sclerotherapy

INTRODUCTION

Infantile Haemangioma (IH) is a benign vascular tumor characterized by endothelial cell proliferation of blood vessels in skin or visceral organs. It appears within the first 2 weeks of life and then generally lesion enlarges in what is called proliferative phase and overall growth by 9 months age. after that there is involution phase which is period of total or partial regression occurring as late as 5 to 10 years of age **(1).** IH present shortly after birth most often as well-demarcated, flat, and erythematous red patches. Haemangiomas follow a predictable course with three distinct developmental phases: proliferation, plateau, and involution. During proliferation, rapid growth can lead to exhaustion of blood supply with resulting ischemia, necrosis, ulceration, and bleeding **(2).** Infantile haemangiomas have been sub classified based on their range of presentations. They are categorized based on depth of tissue involvement. Another

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classification is made by their patterns of distribution as either focal, multifocal, segmental, or indeterminate. Sub-classification of these lesions is imperative to make appropriate treatment decisions **(3).** The cause of IH is unknown and their pathology is not clear. Management of IH includes many options. It can be treated medically by beta blockers. Injection of sclerotherapy is a recent method of management of haemangioma by selective injection of sclerosing agent in feeding vessels causing marked decrease in lesion vascularity and size **(4)**. Surgical excision has great rule also in management of IH especially in involution phase and in superficial lesions not invading deep structures **(5).**

The present study aimed to detect the best time of surgical intervention in management of infantile hemangioma.

PATIENTS & METHODS

A prospective clinical trial study was included in carried out in Zagazig University Hospitals. Approval was obtained from Institutional Review Board (IRB) Zagazig University. Written informed consents were obtained from all patients.The work has been carried out in accordance World Medical Association (Declaration of Helsinki) for studies involving humans before prospective collection of patient's data and after informed consent was obtained from patients.

Inclusion criteria:

Patients aged up to 10 years old with infantile haemangioma disfiguring or complaining.

Exclusion criteria:

Patients older than 10 years old with other congenital anomalies, patients with cardiac disease and patients submitted to previous method of treatment.

Operational design:

Patients with infantile haemangioma fullfiling inclusion criteria were evaluated and received medical treatment with betablockers if not complicated and followed up every 2 months for 6 months for possibility of surgical intervention.

All patients were submitted to complete history taking including demographic data as age and sex. The presenting complaint of the parents as regard onset, course, duration since presentation, possible complications or disfigurement. Past history of previous operations and chronic disease (e.g. cardiac diseases,liver diseases, respiratory disorders). Full general and local examination as general examination for vital signs and other systems to assess fitness for surgery and anesthesia. Local examination for the swelling regarding site, size, shape, multiplicity, consistency, pulsating or not, extent, volume, associated lesions or birthmarks.

Investigations:

Duplex ultrasound to confirm diagnosis and to detect if it is high or low flow. Routine laboratory investigations including: CBC, coagulation profile, kidney function test, liver function test.

Medical treatment:

Propranolol as medical treatment is givenin non complicated cases as follows:

1) Oral suspension produced by dissolving10 mg tablet in 5ml. of water.
2) Dosing strategy:1mg/kg/day for1week,then increased to 2-3 mg/kg/day. Target daily dose administered as 3 divided doses.

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3) Complicated cases or cases that are resistant to medical treatment are subjected to the interventions

Injection sclerotherapy:
Treatment by this method was mainly duplex guided sclerotherapy sessions according to the case. During the follow up, we evaluated symptomatic improvement regarding the decrease in pain and lesion size and obliteration of vascular channels on imaging. The end point was successful reduction in size or complete obliteration of vascular channels. Surgery may be needed as a final stage. Polidocanol was used in concentration of 1-3% according to the size and depth of vascular channels, and acted as a sclerosing agent including damage of vascular endothelial cells, with formation of dense platelet aggregation, and the vessels were replaced by fibrous connective tissue.

Surgical excision:
Complicated cases or resistant to medical treatment are subjected to surgical excision under general anaesthia and after consultation of the parents.

Follow up:
All patients were followed up for 6 months regarding size, shape, duplex ultrasound assessment, presence of complications as bleeding, ulceration, infection or compression on any vital structure.

Statistical analysis:
Data entered and analyzed using Microsoft Excel software. Data were then imported into Statistical Package for the Social Sciences (SPSS version 20.0) software. According to the type of data qualitative represent as number and percentage, quantitative continues group represent by mean ± SD, the following tests were used to test differences for significance; difference and association of qualitative variable by Chi square test(X2). Differences between quantitative independent groups, paired by paired t. P value was set at<0.05 for significant results &<0.001 for high significant result.

RESULTS
The present study showed age was distributed as 2.95±1.57 with minimum 1 and maximum 6 years regard sex distribution male represent 54.2% of studied group and female represent 45.8% **(Table 1)**. Major site was forehead with 25% followed by back & scalp each with16.7% then abdomen, abdomen & back and Lip with 8.3% each and finally arm, buttocks, chest wall and face (one case each) with percentage of 4.2%, regard shape majority were round with 58.3% then irregular 25% and finally oval with 16.7%, regard consistency the majority were soft with 91.7% and 8.3% were firm and majority were solitary 83.3% and only 16.7% (4 cases) were multiple **(Table 2)**.
Regard outcome the majority were good with 37.5% then excellent with 29.2% then bad 16.7% and fair &very good 8.3% **(Figure 1)**. Bleeding reported at 4 cases with 16.7% ulceration and infection 8.3% **(Table 3)**. Medical management was in 66.7% and surgical in 33.3% (8 cases) 6 cases were excision and 2 cases had injection (one of them need excision),finally combined were in 29.2% **(Figure 2)**. There was significant decrease at all time from pre and 2 month (P=0.049*)and from pre to 4

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month (P=0.004*) and from pre to 6 month (P=0.002*) **(Table 4)**. Regarding association between basic demographic and hemangiomas characters and type of management, large size of hemangioma, female gender, abdomen ,back, buttocks and face site were significantly associated with surgical intervention also irregular shape, firm consistency and multiple lesion were significantly associated with surgical **(Table 5)**. Surgical management significantly associated with better outcome **(Table 6)**.

Table (1): Age and sex distribution among studied group (N=24):

		Age/years	
Mean±SD		2.95±1.57	
Median(Range)		3.0(1-6)	
		N	%
Sex	Female	11	45.8
	Male	13	54.2
	Total	24	100.0

Table (2): Hemangioma characters distribution among studied group

		N	%
Site	Abdomen	2	8.3
	Abdomen&Back	2	8.3
	Arm	1	4.2
	Back	4	16.7
	Buttocks	1	4.2
	Chestwall	1	4.2
	Face	1	4.2
	Forehead	6	25.0
	Lip	2	8.3
	Scalp	4	16.7
Shape	Irregular	6	25.0
	Oval	4	16.7
	Round	14	58.3
Consistency	Firm	2	8.3
	Soft	22	91.7
Numberlesions	Multiple	4	16.7
	Solitary	20	83.3
	Total	24	100.0

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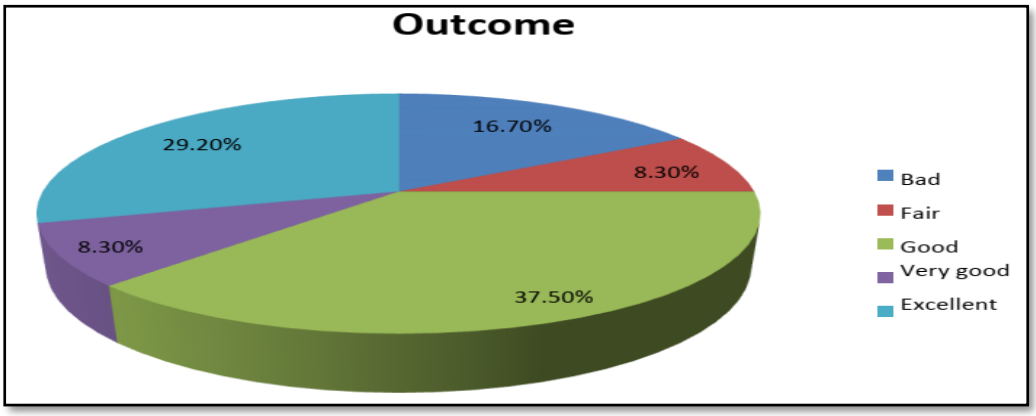

Figure (1):Outcome distribution among studied group (N=24)

Table(3):Complication distribution among studied group:

		N	%
Bleeding	No	20	83.3
	Yes	4	16.7
Ulceration	No	22	91.7
	Yes	2	8.3
Infection	No	22	91.7
	Yes	2	8.3
	Total	24	100.0

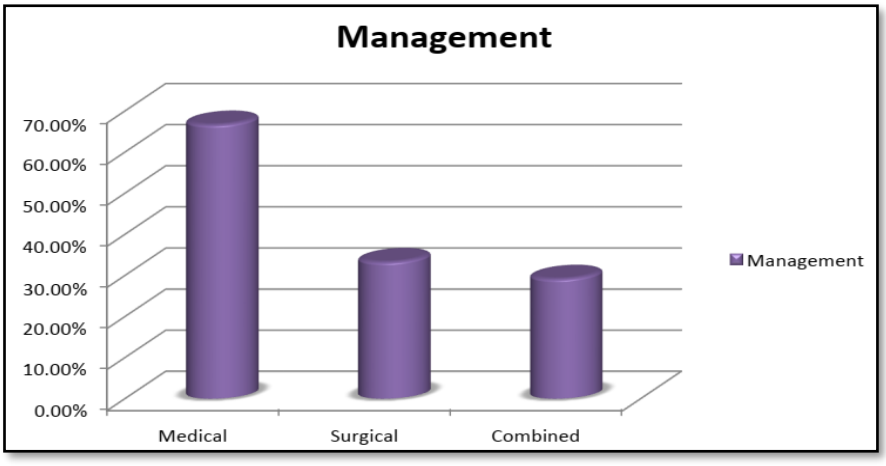

Figure (2):Management distribution among studied group

Table (4): Size of Hemangioma distribution at pre and follow up times

	Size pre	Size 2 months	Size 4 months	Size 6 months
Mean±SD	6.93±2.31	5.61±1.75	4.25±1.53	3.82±1.25
Median(Range)	6.0(2-16)	5.0(2-10.5)	4.0(2-9)	3.75(1-8)

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Table (5): Association between basic demographic and hemangiomas characters and type of management

			Medical	Surgical	P
Age			2.68±0.98	3.51±1.33	0.241
Sizepre			5.65±1.88	9.54±3.12	0.006*
Sex	Female	N	3	8	
		%	18.8%	100.0%	
	Male	N	13	0	0.00**
		%	81.2%	0.0%	
Site	Abdomen	N	0	2	
		%	0.0%	25.0%	
	Abdomen&Back	N	0	2	
		%	0.0%	25.0%	
	Arm	N	1	0	
		%	6.2%	0.0%	
	Back	N	4	0	
		%	25.0%	0.0%	
	Buttocks	N	0	1	
		%	0.0%	12.5%	
	Chestwall	N	1	0	0.035*
		%	6.2%	0.0%	
	Face	N	0	1	
		%	0.0%	12.5%	
	Forehead	N	4	2	
		%	25.0%	25.0%	
	Lip	N	2	0	
		%	12.5%	0.0%	
	Scalp	N	4	0	
		%	25.0%	0.0%	
Shape	Irregular	N	0	6	
		%	0.0%	75.0%	
	Oval	N	4	0	0.00**
		%	25.0%	0.0%	
	Round	N	12	2	
		%	75.0%	25.0%	
Consistency	Firm	N	0	2	
		%	0.0%	25.0%	
	Soft	N	16	6	0.037*
		%	100.0%	75.0%	
Numberoflesions	Multiple	N	0	4	
		%	0.0%	50.0%	
	Solitary	N	16	4	0.002*
		%	100.0%	50.0%	
Total		N	16	8	
		%	100.0%	100.0%	

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Table (6):Association between outcome and type of management

			Management		P
			Medical	Surgical	
Outcome	Bad	N	4	0	0.00**
		%	25.0%	0.0%	
	Fair	N	2	0	
		%	12.5%	0.0%	
	Good	N	9	0	
		%	56.2%	0.0%	
	Verygood	N	1	1	
		%	6.2%	12.5%	
	Excellent	N	0	7	
		%	0.0%	87.5%	
Total		N	16	8	
		%	100.0%	100.0%	

DISCUSSION

Infantile hemangioma (IH) is considered by many clinicians to be a non-medical and non-surgical entity, as spontaneous involution often leads to a favorable outcome. When indicated, medical treatments as topical, systemic and intra-lesional steroids. Pulse dye laser therapy is commonly used for treatment, especially in cases with ulceration or superficial teleangiectasias **(6).**

Surgical treatment may definitively interfere in patient appearance, and this scenario strengthens the elaboration of precise indications for surgery and a comprehensive protocol for its approach **(7).** Although attempts have been made to broadly stratify lesions as low-or high risk based on location, size, and functional impairment with corresponding options for management, controversy continues to exist on the role of surgery for the treatment of IH **(5)**. There are few large studies detailing surgical outcomes, long-term follow-up, and complications after resection **(8).**

In our study, age was distributed as 2.95±1.57 and males represent 54.2% of studied group. While in the studymade by **Daramola et al.(9)** included 1012 patients diagnosed of IH were identified. Of these, 92 patients (9.1%)underwent surgical excision. Female to male ratio was 3:1. Mean and medianages were 36 months and 25 months respectively.

In our study, major site of IH was forehead with 25% followed by back & scalp each with16.7% then abdomen, abdomen & back and lips with 8.3; and arm, buttocks, chest wall and face (one case each) with percentage of 4.2%.

In **Kridin et al. (10)** revealed the leading involved site was the face (71.4%),followed by the limbs (21.4%), the trunk (21.4%), and the genital mucosa (n =1;3.6%). Several patients had simultaneous involvement of more than one anatomical location which is comparable to our study.

Our study agree with **Daramola et al.(9)** studied sixty-three lesions (67%) that were located on the head and neck, with the most common location being the forehead/brow region (37patients), followed by the lip and philtrum (24patients).The average head and neck lesion size was 4.9 cm^2. Out of the 14 cases requiring multiple excisions reported, 12 patients had solitary facial IH, and the primary indication for their additional surgery was completion of excision.

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Moreover, bleeding, visual impairment, feeding impairment, auditory impairment, nasal deformities, congestive heart failure which may occur with large IHs, as a result of arterio-venous shunting, airway obstruction **(11).**

In our study, medical management using propranolol was in 66.7% and surgical in 33.3% (8 cases) 6 cases were excision and 2 cases had injection (one of them need excision) and combined were in 29.2%. This can be compared to a study done by **Daramola et al.(9)** revealed 92 patients (9.1%) underwent surgical excision on a total of 94 lesions with 111 unique visits to the operating room.14 out of 92 patients (15.0%) required a total of 34 surgeries (including first surgery) for completion of excision, treatment of a complication, and surgical site revision. There was no difference between the size of lesions requiring one surgical excision and those that required multiple surgeries.

Demiri et al.(12) reported outcomes as excellent, very good, and good with the majority of results described as excellent or very good. In a similar fashion, **Tian et al.(13)** collected data from 31 IH patients presenting with 32lesions in the post proliferative phase. They were treated with propranolol at adose of 2 mg/kg per day for an average duration of 10.1 months. The clinical outcomes included 17(53.1%) are excellent responses 8(25.0%) good responses and 7 (21.9%) moderate responses, while recurrence was observed in 3 (9.4%) patients.

CONCLUSION

Surgical procedures are generally reserved for complicated Infantile hemangiomas (IH) in which conservative medical therapy is ineffective, not tolerated or contraindicated.

Additionally, surgery is considered in patients with acute complications such as ulceration or hemorrhage.

Finally surgery may be considered when there are likely to be significant functional or cosmetic deficits despite medical therapy or when these deficits might be more severe with the prolonged treatment time frame associated with medical options.

We recommend further studies including larger number of patients and long-term follow up to detect the best time of surgical intervention in management of infantile hemangioma.

No conflict of interest.

REFERENCES

1- **Hoeger, H., Colmenero, I. (2014)**. Vascular tumours in infants. Part I: benign vascular tumours other than infantile haemangioma. British Journal of Dermatology, 171(3), 466-473.
2- **Rommer, E., Anselmo, D. M., Stanley, P., Arkader, A., & Panossian, A. (2013).** Vascular anomalies of the extremities. Current Orthopaedic Practice, 24(3), 244-252.
3- **Buckmiller, L. M., Richter, G. T., & Suen, J. Y. (2010).** Diagnosis and management of hemangiomas and vascular malformations of the head and neck. Oral diseases, 16(5), 405-418.
4- **Mattassi, R., Loose, D. A., & Vaghi, M. (Eds.). (2015).** Hemangiomas and vascular malformations: an atlas of diagnosis and treatment. springer.
5- **Wildgruber, M., Sadick, M., Müller-Wille, R., & Wohlgemuth, W. A. (2019).** Vascular tumors in infants and adolescents. Insights into imaging, 10(1), 1-14.

ISSN:0975-3583,0976-2833 VOL13,ISSUE 01,2022

6- **Wong, M. Y. Y., Yau, G. S. K., Lee, J. W. Y., Chu, A. T. K., & Tam, V. T. Y. (2014).** Management of a large periocular infantile hemangioma with oral propranolol and surgical excision: A Case Report and Review. J Pregnancy Child Health, 1, 103.

7- Chen, Z. Y., Wang, Q. N., Zhu, Y. H., Zhou, L. Y., Xu, T., He, Z. Y., & Yang, Y. (2019). Progress in the treatment of infantile hemangioma. Annals of translational medicine, 7(22).

8- **Yu, Z., Cai, R., Chang, L., Qiu, Y., Chen, X., Chen, Q., Lin, X. (2019).** Clinical and radiological outcomes of infantile hemangioma treated with oral propranolol: A long-term follow up study. The Journal of dermatology, 46(5), 376-382.

9- **Daramola, O., Chun, H., Nash, J., Drolet, A., North, E., Jensen, N., Kerschner, E. (2011).** Surgical treatment of infantile hemangioma in a multidisciplinary vascular anomalies clinic. International journal of pediatric otorhinolaryngology, 75(10), 1271-1274.

10- **Kridin, K., Pam, N., Bergman, R., Khamaysi, Z. (2020).** Oral propranolol administration is effective for infantile hemangioma in late infancy: A retrospective cohort study. Dermatologic therapy, 33(3), e13331.

11- **Darrow, H., Greene, K., Mancini, J., Nopper, J., Section On Dermatology, Section On Otolaryngology–Head & Neck Surgery, And Section On Plastic Surgery (2015).** Diagnosis and management of infantile hemangioma: executive summary. Pediatrics, 136(4), 786-791.

12- **Demiris, G., Finkelstein, M., Speedie, M. (2001).** Considerations for the design of a Web-based clinical monitoring and educational system for elderly patients. Journal of the American Medical Informatics Association, 8(5), 468-472.

13- **Tian, Y., Xu, D. P., Tong, S., Xi, S. L., Liu, Z. M., Wang, X. K. (2016).** Oral propranolol for the treatment of infantile hemangiomas in the post-proliferative phase: A-single center retrospective study of 31 cases. Journal of Oral and Maxillofacial Surgery, 74(8), 1623-1629.