Original Article

Cystic Hygroma in Children: Comparison Between Sclerosing Treatment With Bleomycin Vs Surgical Resection

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ABSTRACT

Objective: To compare the outcome of sclerotherapy with bleomycin versus surgical resection in children in cystic hygroma.

Study Design: Comparative/Observational

Place and Duration: This study was conducted at the Department of Paediatric Surgery, Bolan Medical Complex Hospital Quetta from 1st January 2019 to 30th June 2019.

Materials and Methods: Forty patients of both genders with ages 1 month to 15 years presented with cystic hygroma were included in this study. Patients detailed demographic were recorded after written consent from parents/guardians. Patients were divided in to two groups' i.e. Group A consist of 20 patients and received sclerotherapy with intra-lesional bleomycin and Group B consist of 20 patients received surgical excision. Outcomes were analyzed at post-procedure 3 and 6 months post-procedure and compare the findings between both groups.

Results: There were significant difference observed between both groups regarding age, gender and site of lymphangioma p-value >0.05. In Group A 15 (75%) patients showed excellent results, 3 (15%) patients showed good results and 2 (10%) showed poor results. In Group B 9 pts (65%) showed excellent results, 5 (20%) showed good results and 6(15%) showed poor results. At final follow-up there were 2 patients with recurrence in Group A while in Group B 5 (10%) patients had recurrence,

Conclusion: Sclerosing treatment with bleomycin is safe and effective treatment modality with no recurrence as compared to surgical resection.

Keywords: Cystic hygroma, Sclerotherapy, Bleomycin, Surgery, Recurrence

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INTRODUCTION

Lymphangiomas are benign hamartomatous lymphatic tumors, also referred to as congenital deformities of the lymphatic structures. The most common and frequent type of lymphangioma is cystic hygroma which may compose of one or more macrocytic lesions with reduced communication to the lymphatic channels.[1] They are slow growing tumors and may manifest in any part of the body or anywhere in the soft tissues.

The most commonly affected sites are the head and neck, and also the mediastinum and axilla[2]. These tumors most often occur in children, although they may occur in adults as well.

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They are also often evident at birth in 65% and may present by two years of age in 80-90% of the cases. The reported incidence of lymphangiomas is 1.5 to 2.8 per 1000, and it is not specific to any gender or race.[2] Inflammation and cosmetic defects are the baseline symptoms. In the neck area, they may lead to compression symptoms when they compress important surrounding structures and may cause respiratory problems, dysphagia, and compression of nerves.[3]

There are different modes of treatment for cystic hygromas. These therapies include radiotherapy, incision and drainage, and surgical excision (which is the mainstay of therapy).[2,4-6] However, these procedures have variable findings which investigators find unsatisfactory. In many patients, a complete surgical resection is often not possible because of the nature of the lesion which encircles the crucial neurovascular system and also infiltrates across tissue planes.[2] Post-surgical recurrence of tumors and injury to nerves are commonly observed complications, in addition to other issues such as disfigurement, and dreadful scar formation.[1,2]

In recent times, intralesional sclerotherapy has become increasingly accepted as an effective method for the treatment of the children with lymphangiomas.[1,2,4] It involves the use of a sclerosing agent which irritates the endothelial lining of the lymphangioma leading to

swelling, involution, and fibrosis. In the past, boiling water, 50% dextrose water, hypertonic saline, or absolute alcohol have been used with results that have not been very encouraging.[2] Many centers are beginning to use sclerosing agents like Bleomycin, acetic acid, OK-432, and Doxycycline as first-line therapy with satisfactory results.[1,3,7,8] Unlike surgical excision, this modality of treatment is particularly useful for lesions enveloping vital structures.[8] The present study was conducted to compare the effectiveness and safety of sclerotherapy and surgical excision in children with cystic hygroma.

MATERIALS AND METHODS

This observational study was conducted at Bolan Medical Complex Hospital Quetta from 1st January 2019 to 30th June 2019. A total 40 patients of both genders with ages 1 month to 15 years presented with cystic hygroma were included in this study. Patients detailed demographic including age, sex, and site of lymphangioma were recorded after taking written consent from parents/guardians. Patients with recurrence, already on sclerotherapy and ages above 15 years were excluded from this study. All the patients were divided in to two groups' i.e. Group A consist of 20 patients and received sclerotherapy intra-lesionally bleomycin with a dose of 0.5mg/kg of body weight and number of session 1 to 4 per patients. Patients were admitted for 24 hours after each session. Maximum 4 sessions was given to patients monthly. Group B consist of 20 patients received surgical excision of cystic hygroma. Post-procedural complications were recorded. Outcomes in term of excellent (Complete resolution), good (>50% resolution) and poor (<50% resolution) results were examined. Recurrence rate was examined at final follow-up. Follow-up was taken at 3 and 6 months post-procedure. Ultrasonography and X-ray was done pre and post-operatively to analyze the outcomes. Data was analyzed by SPSS 24.0. Chi-square and student t' test was used to compare the outcomes between both groups. P-value <0.05 was considered as statistically significant.

RESULTS

There were 11 (55%) male and 9 (45%) were females in Group A and in Group B 12 (60%) patients were males and 8 (40%) patients were females. In group A majority 75% of patients were ages <5 years and 25% patients had ages above 5 years. In Group B 80% patients were ages ≤ 5 years and 20% patients had ages above 5 years. According to the site of lymphangioma, in Group A 60% patients had neck. 15% had axilla, 15% had face and 10% had trunk site and in Group B 55% patients had neck, 20% had axilla, 15% had face and 10% patients had trunk lymphangioma. There was no significant difference observed regarding age, sex and site of lymphangioma between both groups with p-value >0.05 (Table 1).

In Group A patients 1 (5%) patients had received two sessions of bleomycin, 4 (20%) had received three sessions and 15 (75%) had received 4 doses of bleomycin (Table 2). According to the post-procedural complications we found no patient had wound infection in Group A patients while in Group B 2 (10%) patients had developed wound infection and 8 (10%) patients had recurrence.

Table No.1: Baseline details of all the patients

Variable	Group A (n=20)	Group B (n=20)	P-value
Gender			
Male	11 (55%)	12 (60%)	>0.05
Female	9 (45%)	8 (40%)	
Age (years)			
<5	15 (75%)	16 (80%)	>0.05
>5	5 (25%)	4 (20%)	
Site of lymphangioma			
Neck	12 (60%)	11 (55%)	>0.05
Axilla	3 (15%)	4 (20%)	
Face	3 (15%)	3 (15%)	
Trunk	2 (10%)	2 (10%)	

Table No.2: Session-wise distribution in patients receive bleomycin

Variables	No.	%
One Session	-	-
Two Sessions	1	5.0
Three Sessions	4	20.0
Four Sessions	15	75.0

Table No.3: Post-operative complications between both groups

Complication	Group A (n=20)	Group B (n=20)	P-value
Wound Infection			
Yes	-	2 (10%)	>0.001
No	20 (100%)	18 (90%)	
Recurrence			
Yes	-	8 (10%)	>0.001
No	20 (100%)	18 (90%)	

Table No.4: Final outcomes between both groups

Variables	Group A (n=20)	Group B (n=20)	P-value
Excellent	15 (75%)	9 (45%)	>0.05
Good	3 (15%)	5 (25%)	>0.05
Poor	2 (10%)	6 (30)	>0.05

There was a significant difference between both groups (p=<0.001). According to the resolution we found significant difference between both groups in term of complete resolution with p-value >0.05. In Group A 15 (75%) patients showed excellent results, 3 (15%) patients showed good results and 2 (10%) showed poor

results. In Group B 9(65%) showed excellent results, 5 (20%) showed good results and 6 (15%) showed poor results (Table 4).

DISCUSSION

Cystic hygroma is one of the critical disorder among children and the incidence rate is quite high in children with ages up to 5 years. Many of treatment modalities such as sclerotherapy with bleomycin and OK-432 and the surgical excision have been applied for this benign disorder. In these modalities sclerotherapy technique is considered as much safer and effective than surgical management due to high rate of wound infection and recurrence rate.[9,10] The present study was conducted aimed to examine the outcomes of sclerotherapy with bleomycin and surgical excision in children with cystic hygroma. In this regard 40 patients of both genders were enrolled and divided in to two groups. We found that male patients were high in numbers in both groups A and B 55% and 60% as compared to females 45% and 40%. In group A majority 75% of patients were ages <5 years and 25% patients had ages above 5 years. In Group B 80% patients were ages $\leq$ 5 years and 20% patients had ages above 5 years. A study conducted by Mustafa et al[11] regarding outcomes of intralesional bleomycin for cystic hygroma in children, in which they reported that male patients was high in numbers 66.7% as compared to females and the mean age of patients was 2.36 ±2.8 years.

In present study according to the site of lymphangioma, in Group A 60% patients had neck. 15% had axilla, 15% had face and 10% had trunk site and in Group B 55% patients had neck, 20% had axilla, 15% had face and 10% patients had trunk lymphangioma. There was no significant difference observed regarding age, sex and site of lymphangioma between both groups with p-value >0.05. A study by Fiaz et al[12] reported that neck was the commonest site of lymphangioma found in 63.3% followed by axilla, face and trunk 13.3%, 13.3% and 10%.

In the present study we found that patients who were received sclerotherapy, 1 (5%) patients had received two sessions of bleomycin, 4 (20%) had received three sessions and 15 (75%) had received 4 doses of bleomycin. These results were similar to many of other studies in which mostly patients were received 3 to 4 session of bleomycin for complete resolution.[13,14]

According to the post-procedural complications, we found no patient had wound infection in Group A patients while in Group B 2 (10%) patients had developed wound infection and 5 (10%) patients had recurrence in group B . while in group A 2 patients have recurrence .There was a significant difference between both groups (p=<0.001). These results were similar to many of previous studies in which surgical excision had high rate of wound infection 5 to 20% and recurrence rate 10 to 30% as compared to sclerotherapy.[15,16] We observed that surgical excision needs much per-operative care as compared to sclerotherapy.

This study showed that significant difference between both groups in term of complete resolution with p-value >0.05. In Group A 15 (75%) patients showed excellent results, 3 (15%) patients showed good results and 2 (10%) showed poor results. In Group B 09 (65%) showed excellent results, 04(20%) showed good results and 06 (15%) showed poor results. These results were comparable to several previous studies.[17-20]

CONCLUSION

Sclerosing treatment with bleomycin for cystic hygroma in children is safe and effective treatment modality with no recurrence and wound infection as compared to surgical resection. Also we observe significant difference between both groups in term of complete resolution, good and poor resolution.

Author's Contribution:

Concept & Design of Study:	Mohammad Dawood Khan
Drafting:	Habib ullah Mandokhail
Data Analysis:	Asmat Ullah Kaker
Revisiting Critically:	Mohammad Dawood Khan, Habib ullah Mandokhail
Final Approval of version:	Mohammad Dawood Khan

Conflict of Interest: The study has no conflict of interest to declare by any author.

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