

# Comparative Analysis of Sclerotherapy with Bleomycin Versus Surgical Resection for Cystic Hygroma in Children

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## ABSTRACT

**Objective:** Main purpose of this study is to compare the outcomes of sclerotherapy with bleomycin versus surgical resection in children with cystic hygroma.

**Study Design:** Comparative/Observational study

**Place and Duration:** This observational study was conducted at Children Surgical Hospital, Sialkot for six months during the period from July 2020 to December 2020.

**Methods:** Total 50 children of both genders were presented. Children were aged between 2-12 years. All the patients had cystic hygroma were included. Informed written consent was taken from parents of children for detailed demographics. Patients were equally divided into two groups, group I had 25 patients and received sclerotherapy with bleomycin and group II received surgical resection among 25 cases. Post-treatment outcomes were assessed and compared in terms of efficacy and recurrence among both groups. SPSS 22.0 version was used to analyze complete data.

**Results:** Majority were males in group I and II (68% and 64%) and females were 8 (32%) in group I and 9 (36%) in group II. In group I 18 (72% cases) were <6years of age and in group II 19 (76%) children were < 6years of age. Neck and axilla was the most effected site of cystic hygroma followed by face and trunk. We found that effectiveness of sclerotherapy with bleomycin was higher among 21 (84%) cases as compared to surgical resection 19 (76%). Recurrence rate was higher in group II 5 (20%) as compared to group I 3 (12%). Wound infection was lower in group I 1 (4%) and in group II we found 3 (12%).

**Conclusion:** In this research we concluded that use of sclerotherapy with bleomycin is an effective and safe treatment for cystic hygroma as compared to surgical resection in terms of less recurrence rate, wound infection and higher number of good results.

**Keywords:** Cystic hygroma, Children, Sclerotherapy, Bleomycin, Surgical Resection

## INTRODUCTION

In addition to being known as water-tumor, lymphangioma, and cystic hygroma, cystic hygroma is a benign abnormality of the lymphatic vessels that usually forms when the lymphatic system fails to connect with the normal jugular vein. Axilla, cervico-facial regions and axilla are all possible sites of involvement. In 1828, Reden Backer published the first report of lymphangioma, a soft tissue tumour with a controversial pathogenesis. In 1834, Wernker published the first account of "cystic hygroma," a soft tissue tumour with an unknown pathogenesis. [2] When there is vigorous lymphatic development, Lymphangioma is often regarded as a paediatric disease. Cystic hygromas are single or multiple cysts that are most commonly encountered in the neck region of the human body. Cystic hygroma can occur as a birth condition (congenital) or can develop at any point in a person's life at any point in their lives. [3]

With a recurrence rate of 21 percent, they are locally aggressive, benign lesions that are challenging to control since the tumour recurrence after surgery makes it tough to manage the condition.

In most cases, the proliferation of tiny lymphatic arteries with intervening fibrous tissue is what distinguishes cystic hygroma from other types of tumours. When it manifests itself in adulthood, it is extremely unusual and the aetiology is unknown, but trauma and upper respiratory tract infection have both been cited as potential triggers.

[4] These abnormalities are most typically found in the head and neck region, while they have been reported in a variety of other anatomical regions as well. In the English language literature, there have only been a few hundred fifty instances of adult cervicofacial cystic hygroma, and the best way to treat these lesions is still up in the air, according to some experts. A final diagnosis is usually made based on postoperative histology in adults because they are believed to be more difficult to diagnose than children. [4]

Despite the fact that CH can affect any anatomic subsite in the human body [5-7], the majority of these lesions are found in the head and neck region (75 percent), which is likely due to the abundance of lymphatics in this region. Cystic Hygroma (CH) typically affects children under 2 years of age (80–90 percent), with an incidence rate of 1.2–2.8 per 100,000 newborns [8,9]. Both males and females are equally impacted by CH, which indicates that it has no gender preference [10, 11].

Although complete surgical removal is a valuable treatment option [12], it is not always possible since the lesion tends to expand along essential structures. As a result, in some individuals, complete surgical removal is not possible. Many adjuvant therapy have been demonstrated to be effective in the treatment of recurring or inaccessible lesions, but they are not routinely utilised. Another successful therapy option for sclerosis is a chemical medication [13]. Only a few cases of adult cervical CH have been reported in the literature to date, and determining the

most effective treatment strategy remains a difficult task. Patients are frequently misdiagnosed prior to surgery in adults because preoperative diagnosis is challenging. According to the example reported here, patients with CH are frequently misinterpreted as having brachial cleft cysts on imaging. Only after histological inspection of the surgical specimen can a definitive diagnosis be made.

Purpose of this study was to compare the outcomes of sclerotherapy with blooming versus surgical resection in children with cystic hygroma.

**MATERIAL AND METHODS**

This observational study was conducted at Children Surgical Hospital, Sialkot for six months during the period from July 2020 to December 2020. A total 50 patients of both genders with ages 2 to 12 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 12 years were excluded from this study.

All the patients were divided in to two groups' i.e. Group I consisted of 25 patients who got sclerotherapy with bleomycin at a dose of 0.3mg/kg of body weight and a total of 1 to 4 sessions per patient. After each session, patients were required to remain in the facility for 24 hours. Patients were only allowed to have a maximum of four sessions each month. Group II is made up of 25 individuals who underwent surgical removal of cystic hygroma tumours. There were no post-procedural problems that were documented. The outcomes were categorised into three categories: excellent (complete resolution), good (>50 percent resolution), and bad (less than 50 percent resolution) results. At the conclusion of the study, the rate of recurrence was determined. Three and six months after the procedure, a follow-up was performed. Pre- and post-operative ultrasonography and X-rays were performed in order to evaluate the outcomes. Data was analyzed by SPSS 22.0. 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**

Table 1: Baseline detailed demographics of enrolled cases

Variables	Group I	Group II
Age		
<6years	18 (72%)	19 (76%)
>6years	7 (28%)	6 (24%)
Gender		
Male	17 (68%)	16 (64%)
Female	8 (32%)	9 (36%)
Site of hygroma		
Neck	14 (56%)	13 (52%)
Axilla	5 (20%)	6 (24%)
Face	4 (16%)	3 (12%)
Trunk	2 (4%)	3 (12%)

Majority were males in group I and II (68% and 64%) and females were 8 (32%) in group I and 9 (36%) in group II. In group I 18 (72%) cases were <6years of age and in group II 19 (76%) children were < 6 years of age. Neck and

axilla was the most effected site of cystic hygroma followed by face and trunk.(table 1)

We found that effectiveness of sclerotherapy with blooming was higher among 21 (84%) cases as compared to surgical resection 19 (76%).(table 2)

Table 2: Post treatment comparison of efficacy among both groups

Variables	Group I (n=25)	Group II (n=25)	P-value
Excellent	17 (68%)	14 (56%)	<0.05
Good	4 (16%)	5 (20)	<0.05
Poor	4 (16)	6 (24)	<0.05

Recurrence rate was higher in group II 5 (20%) as compared to group I 3 (12%). Wound infection was lower in group I 1 (4%) and in group II we found 3 (12%)

Table 3

Variables	Group I (n=25)	Group II (n=25)	P-value
Recurrence			<0.001
Yes	3 (12%)	5 (20%)	
No	22 (88%)	20 (80%)	
Wound Infection			<0.001
Yes	1 (4%)	3 (12%)	
No	24 (96%)	22 (88%)	

**DISCUSSION**

Cystic hygroma is one of the most serious conditions that can affect children, and the incidence rate is relatively high in children under the age of five. A variety of treatment options, including sclerotherapy with bleomycin and OK-432, as well as surgical excision, have been used to treat this benign condition. Because of the high likelihood of wound infection and recurrence in these modalities, sclerotherapy approach is believed to be significantly safer and more successful than surgical management. [14,15] Using radiological imaging to diagnose and plan therapy for large suspected lymphatic malformations is critical for accurate diagnosis and treatment planning. An ultrasound is frequently sufficient and, in many cases, the most appropriate modality in young patients in terms of compliance; it also avoids the need for additional anaesthesia to undertake any additional scanning, such as an MRI or CT scan. If an ultrasound scan does not provide sufficient information, an MRI scan is recommended, and it is superior to a CT scan in most cases.

In this observational/comparative study 50 children were presented. Children were aged between 2-12 years. Patients were equally divided into two groups, group I had 25 patients and received sclerotherapy with bleomycin and group II received surgical resection among 25 cases. Majority were males in group I and II (68% and 64%) and females were 8 (32%) in group I and 9 (36%) in group II. In group I 18 (72%) cases were <6years of age and in group II 19 (76%) children were < 6 years of age. Previously different studies presented same results to our study.[16,17] Neck and axilla was the most effected site of cystic hygroma followed by face and trunk.[3,4] t is likely that hygromas are caused by the sequestration of lymphatic tissue that has kept its ability to proliferate in any of these locations. However, the aetiology of the disease in the adult population is still debated. Some writers believe that adult lymphangioma is caused by the delayed

proliferation of congenital or acquired lymphoid resting as a result of trauma or respiratory infections prior to diagnosis.

In current study we found that effectiveness of sclerotherapy with bleomycin was higher among 21 (84%) cases as compared to surgical resection 19 (76%). The use of imaging guided sclerotherapy with doxycycline [18], doxycycline coupled with sodium tetradecyl sulphate [19], and ethanol insertion [20] in children with lymphatic anomalies have all demonstrated high success rates in these patients. Radiographic resolution rates for nonsurgical treatment of macrocystic lymphatic malformations, such as cystic hygromas, were found to be between 87.5 percent and 95.2 percent in these trials, demonstrating good results in terms of radiographic resolution. Despite the fact that the study had a small number of participants, there were very few issues observed. All studies, on the other hand, indicated limited effectiveness in the treatment of microcystic/simple abnormalities, such as lymphangioma. Additionally, there was no testing for intraoral abnormalities.

Recurrence rate was higher in group II 5 (20%) as compared to group I 3 (12%). Wound infection was lower in group I 1 (4%) and in group II we found 3 (12%). These findings were consistent with many earlier studies in which surgical excision was associated with a high rate of wound infection (5 to 20 percent) and recurrence (10 to 30 percent) as compared to sclerotherapy [21,22], as well as a high rate of recurrence (10 to 30 percent). When comparing surgical excision to sclerotherapy, we discovered that the latter requires significantly more post-operative care.

Cystic hygroma's behaviour during infancy is diverse; sometimes it grows very quickly and causes respiratory trouble, a condition that necessitates urgent suction of much of the contents of cysts and the placement of a tracheostomy tube. It is also possible that the swelling will become irritated as a result of nasopharyngeal infection. In this case, spontaneous regression of cystic hygroma will occur.

## CONCLUSION

In this research we concluded that use of sclerotherapy with bleomycin is an effective and safe treatment for cystic hygroma as compared to surgical resection in terms of less recurrence rate, wound infection and higher number of good results.

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