

# Intralesional Bleomycin for Lymphangioma: An Effective Alternative Non-surgical Therapy

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## Abstract

**Introduction:** Conventionally, lymphangiomas have been managed by surgical excision which range from simple excision to extensive compartmental exenteration. With the advent of the usefulness of sclerotherapy, especially bleomycin, management of extensive lymphangiomas which are hitherto considered inoperable cases has become very comfortable for patient and surgeon.

**Materials and Methods:** A total of 28 patients were included in the study. Neck was the most common site of involvement followed by axilla; groin was involved in two cases. Injection bleomycin was reconstituted and diluted, a dose of 0.5 IU/Kg injected intralesionally after aspirating an equal volume of fluid. Compression bandage was applied for 24 h. Cases were reviewed after 3 weeks for assessing clinical and serological response.

**Results:** Significant reduction of mass was noted in 68% of cases ( $n = 19$ ). Surgery was required in 7% ( $n = 2$ ), complete regression of mass was noted in the remaining 25% ( $n = 7$ ) cases.

**Conclusion:** This modality of treatment may be used safely as primary modality of treatment for select group of patients.

**Key words:** Bleomycin, Lymphangioma, Sclerotherapy

## INTRODUCTION

Lymphangioma is a unilocular or multilocular congenital malformation of lymphatic system occurring in approximately 1 in 6000-12000 births.<sup>[1]</sup> It can lead to morbidity due to cosmesis, compression of adjacent organs infection, hemorrhage, rupture, and sinus formation.<sup>[1,6]</sup> Surgery is the mainstay of treatment but has unacceptable rates of complications and morbidity.

Certain newer modalities of treatment have been studied in the past few years, of which intralesional bleomycin therapy has been proved to be highly effective and safe mode of treatment. We present our series of 28 cases of the lymphangiomas treated at our institute which are primarily treated by intralesional bleomycin sclerotherapy.

## MATERIALS AND METHODS

A total of 28 cases of lymphangiomas were diagnosed and treated between 2015 and 2018 at our Institute Rangaraya Medical College, Kakinada. In all cases, diagnosis was established by clinical features sonological and cytological findings. Computed tomography/magnetic resonance imaging was needed in few cases to assess the extent of lesion, particularly into thorax or abdomen. Bleomycin injection<sup>[2,4,5,10]</sup> was used in all cases at a dose of 0.5 IU/Kg body weight after diluting with normal saline to make the required volume average numbers of injections needed were around three in most cases.

The injection was done in the operation theater under sedation after aspirating an equal quantity of lymphangioma fluid [Figure 1a-c]. Multiple site injections were made as most of them are multiloculated. Response was observed by serial photographs and imaging studies.

## RESULTS

Of 28 cases, there was a significant response in 19 cases (68%), good response was noted in 7 cases (25%), and

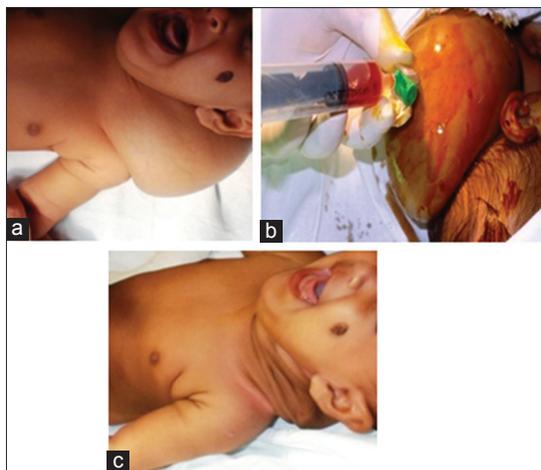
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**Figure 1: (a) Extensive lymphangioma, (b) method of injection, (c) immediate result**

poor response two cases which required surgical excision.

## DISCUSSION

Lymphangiomas are common developmental anomaly of the lymphatic system, the most common site is head and neck, of 28 cases, 18 cases were male and 10 cases were female.

Treatment of lymphangioma is highly challenging. Although surgical excision offers the opportunity for permanent cure by excising the lesion completely, it is practically difficult to achieve this goal by sparing the vital neurovascular structure in many cases.

Recurrences and not so cosmetic outcomes are quite common after surgical procedure in extensive lesions.

In the past two decades, many case series have been reported using sclerosants such as OK-432,<sup>[11,12]</sup> bleomycin, and other sclerosants with great success.<sup>[1]</sup>

Bleomycin is easily available economical and virtually no side effects were reported with intralesional use.

In our study, intralesional bleomycin resulted in significant reduction of size in 68% of case and complete resolution in 25% of cases.

Only side effects noted in our study were initial swelling and signs of local inflammation which settled down with oral paracetamol.

The most dreaded complication of bleomycin is pulmonary toxicity/retroperitoneal fibrosis. This complication has been noted only in subjects who received >400 IU. Hence, the fear of this complication does not arise in the treatment of lymphangioma as the dose used is very small.

## CONCLUSION

Our series concludes that intralesional bleomycin is a very safe and effective method of treating lymphangiomas, thereby avoiding morbid consequences of surgical excision, especially in extensive lesions.

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