ORIGINAL ARTICLE

The Treatment of Neck Lymphangioma with Intralesional Injection of Bleomycin

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Summary

We report our experience with intralesional injection of bleomycin in the treatment of neck lymphangioma. From May 1995 to April 1998, 11 patients aged between 6 to 22 months were treated with intralesional bleomycin injection. Ultrasonography and computed tomography were used to assess and select the cases suitable for sclerotherapy. Patients with lesions encasing the internal jugular vein and the carotid artery were chosen. With the patient under sedation and using ultrasound guidance, the cysts were aspirated and bleomycin was injected at a dose of 0.5mg/kg body weight. The number of procedures varied from 1 to 4 over a period of 8 months to 1 year. Patients were initially followed-up 3 monthly, then 6 monthly and subsequently yearly. In 4 patients, the neck mass was no longer visible (excellent response). In 5 patients, the neck mass had reduced to a size (more than 50% reduction) that was cosmetically acceptable (good response). There were 2 failures (poor response). There were no complications. Our results suggest that intralesional injection of bleomycin can be effectively used to treat selected cases of neck lymphangiomas.

Key Words: Cystic lymphangioma, Sclerotherapy, Bleomycin therapy, Cystic hygroma

Introduction

Lymphangioma is a congenital malformation of the lymphatic vessels. It usually manifests at birth or before the age of 2 years. The neck is the most common site. Some lymphangiomas grow steadily and may cause airway obstruction, feeding difficulties and cosmetic problems. Recurrent inflammation and sudden haemorrhage are potential life threatening complications especially in cervico-mediastinal lesions. For many years surgery was the treatment of choice. However, encasement of adjacent structures such as nerves and vessels may result in nerve injury and other serious complications. Often, total resection is difficult and incomplete excision may lead to lymphorrhea, wound infection and tumour recurrence[1]. Since the late seventies, various types of sclerotherapy have been tried as an alternative to surgical excision. We report our experience with intralesional injection of bleomycin in the treatment of neck lymphangioma.

Materials and Methods

From May 1995 to April 1998, 11 patients with neck lymphangioma aged between 6 to 22 months were treated with intralesional bleomycin injection. The lymphangiomas ranged in size from 3 x 4cm to 10 x 12cm. Ultrasonography and computed tomography were used to assess the suitability for bleomycin therapy. Patients in whom surgical excision was thought to be challenging i.e. where the lesion had encased the internal jugular vein and the carotid artery were chosen.

The clinical indications for treatment were: 6 for cosmetic reasons, 4 for progressive increase in size and 1 for recurrence following surgery.

With the patient under sedation and using ultrasound guidance, the cysts were aspirated with a 23G needle. The intracystic position of the needle was maintained and bleomycin solution (1.5mg/ml water) was injected at a dose of 0.5mg/kg body weight (total maximum dose 5mg/kg). Patients were discharged after 24 hours.

Patients were initially followed-up clinically and with ultrasonography every 3 months until no further intervention is feasible, then 6 monthly until the size of the lesions is stable and subsequently yearly. The patients have been followed-up for a period of between 3 months to 2 years with a mean of about 6 months. The decision to repeat the procedure was based on the amount of reduction in the size of the mass and the presence of cysts that were large enough (more than 1cm) to be aspirated. The number of procedures varied from 1 to 4, over a period of 8 months to 1 year.

Results

In our series, 5 patients had single procedures and 4 patients required additional injections. In 4 patients, there was complete clinical resolution (excellent response) (Fig. 1, 2 and 3). In 5 patients, there was more than 50% regression (good response) and the neck mass had reduced to a size that was cosmetically acceptable. Further procedures could not be performed because there were no further cysts suitable for aspiration. There were 2 failures. Failure was due to inability to aspirate any of the cysts as they were too small.

There were no complications. None of the patients needed subsequent surgery in the limited follow-up period.

Discussion

Various types of sclerosants have been used to treat neck lymphangioma. These include bleomycin solution, bleomycin fat emulsion, doxycyclin, OK-432 and Ethibloc[1-8]. Bleomycin is an antineoplastic antibiotic, doxycyline is a derivative of tetracycline, OK 432 is a product derived from *streptococcus pyogenes* and Ethibloc is an alcoholic solution of zein (corn protein).

The exact mechanism of action of these sclerosants is uncertain. It is thought that the sclerosants cause irritation of the endothelial lining of the cysts[4]. This leads to inflammation and eventual fibrosis.

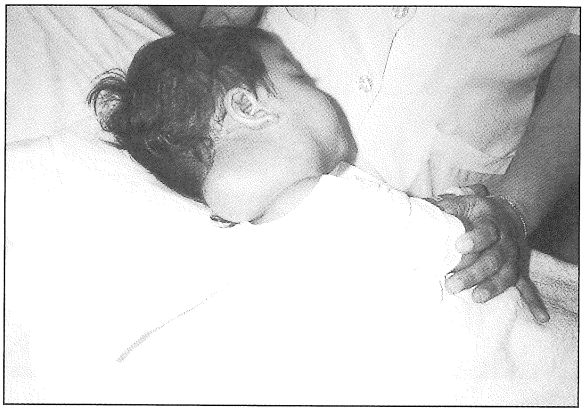

Fig. 1: A cervical lymphangioma measured 10 x 12 cm at presentation in a child aged 22 months. Three cysts were aspirated and 1.5 mg bleomycin solution was injected into each cyst.

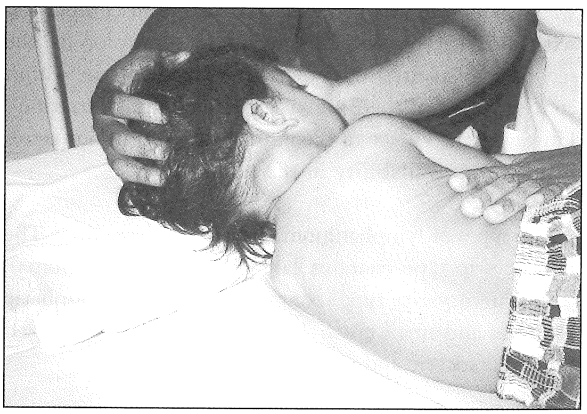

Fig. 2: Three months after the procedure, the mass showed marked reduction in size. The procedure was repeated.

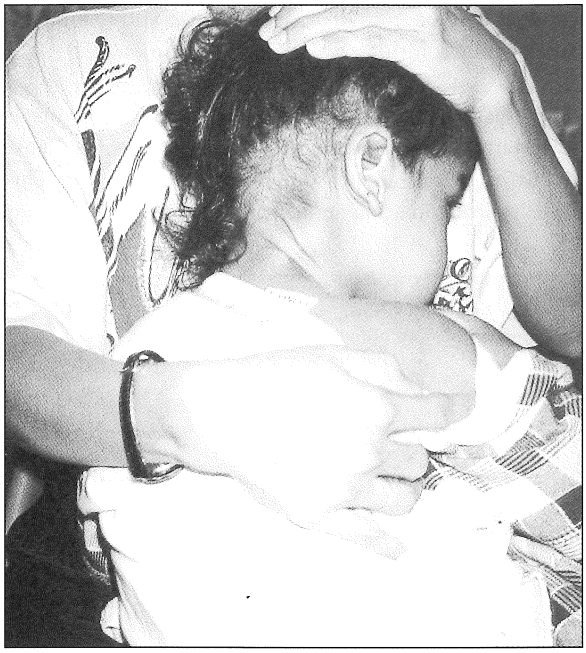

Fig. 3: Three months after the second procedure, there was complete resolution.

The various authors using the different types of sclerosants have obtained variable results: complete resolution were reported in 55% to 64% of cases and satisfactory reduction in size were reported in 36% to 86% of cases[1-7]. Although the response to sclerotherapy was variable, it was generally favourable in comparison with surgical results. Bleomycin fat emulsion is the recommended drug of choice, since it results in a high concentration of the drug at the site of administration[8]. However, the choice of bleomycin solution in our series is due to its availability in the country.

Three types of lymphangioma have been described[6]. The macrocystic type contains large cysts (cystic hygroma). The microcystic type contains abnormal lymphatic channels and small cysts. The majority are of the mixed type: macrocytic and microcystic.

The success rate of sclerotherapy is dependent upon the predominant component of the lymphangioma. The predominantly cystic lesions (cystic hygroma) are the ones that result in an excellent response as in 4 of our cases where there was complete resolution. In the mixed type of lesions, once the bigger cysts have been aspirated leaving the channels and small cysts, further procedure cannot be performed. We had 5 such cases and the lesions reduced to a size that was cosmetically acceptable. Lesions containing predominantly abnormal lymphatic channels or contain small cysts are not suitable for sclerotherapy. We had 2 such cases and we observed that this type of lymphangioma tends to be stable and may not increase in size as the child grows. However, the risk of infection persists in lesions with no or incomplete response and they need careful long-term follow-up.

There were no complications in our series. A similar series using bleomycin solution also did not encounter any complications[1]. Complications such as marked swelling, induration, erythema and pain have been reported with the use of bleomycin fat emulsion[2,3]. However, these complications were not serious. There is a significant risk of airway compromise due to oedema after bleomycin injection in cervico-mediastinal lymphangioma and intralesional injection is contraindicated in such cases[3,8]. It is also advisable to avoid bleomycin sclerotherapy in infants less than 6 months of age. A potential serious complication of using bleomycin is pulmonary fibrosis. However, this risk is dose related. The doses used for sclerotherapy are much lower than those used for oncology i.e. a dose of less than 1mg/kg, administered at not less than 2 week intervals with the total dose limited to 5mg/kg[1]. There is an increased incidence of pulmonary fibrosis with a single dose exceeding 30mg or a total dose of 400mg[3]. Various Japanese series using bleomycin as a sclerosant did not report pulmonary fibrosis as a complication[2,3]. In our series, a longer follow-up period is required to assess our recurrence rate and the long term side effects of bleomycin therapy.

Conclusion

Intralesional bleomycin injection is a safe and effective method of treatment of cystic lymphangioma. It is a favourable alternative to surgical excision in selected cases.

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