Abstract:

Lymphangiomas are benign hamartomatous lymphatic tumours characterized by multiple communicating lymphatic channels and cystic spaces. Cystic lymphangiomas may be seen anywhere in the body but are common in neck, axilla, mediastinum and groin. Treatment options are surgical resection, radiation, sclerotherapy etc but have got high recurrence rate and complications. In the present study we hypothesized that complete surgical resection along with keeping a drain for 2-3 wks, may reduce recurrence rate. This prospective study was performed from July 2008 to June 2013. Total 21 children treated during this period by the technique of complete surgical excision with keeping drain tube in situ for 2 to 3 weeks. Ultrasound performed in all patients, CT scan and Duplex study performed in some patients, histopathology done in all patients. Patients with intra-abdominal lymphangioma not included in this study. Twenty one patients were operated during the study period and average duration of keeping drain tube was 18.57 days. Among the 21 cases 2 (9.52%) patients developed mild infection, 1 (4.76%) patient developed infection with pus formation and 1 (4.76%) patient developed recurrence. Our success in the treatment warrants further verification of potential benefits of keeping drain tube in situ for 2-3 weeks after surgical excision of lymphangiomas.

Key words: Cystic hygroma, excision, keeping drain.

Introduction:

Lymphangiomas are benign hamartomatous lymphatic tumours characterized by multiple communicating lymphatic channels and cystic spaces. Many lymphangiomas involve the skin and subcutaneous tissues and are frequently associated with other type of vascular disorders, particularly venous malformations. Cystic lymphangiomas may be seen anywhere in the body but are common in neck, axilla, mediastinum, groin.

Embryologically these lesions are believed to originate from a pinching off of the developing lymphaticovenous sacs, yielding sequestrations of lymphatic tissue that fail to communicate with the remainder of lymphatic system. It accounts for 5% of all benign tumours in infants and children.

Treatment of choice is surgical resection. Surgical resection has historically had a moderate rate of recurrence of 10%-40% and often requires re-operation. Treatment of recurrent lymphangiomma with radiation, steroids or sclerosing agents has generally been ineffective. Intralesional injection of OK-432, a monoclonal antibody produced by incubation and interaction of low virulence streptococci and penicillin, has an approximately 60% response rate. Intralesional bleomycin has a reported rate of complete resolution in 55% of cases. Complication of bleomycin includes infection, G I disturbances and pulmonary fibrosis.

Although these therapies have shown promise, it is not available at all institutions, making alternative therapy necessary. We hypothesized that complete surgical resection along with keeping a drain for 2-3 weeks, may reduce recurrence rate. The purpose of this study is to evaluate the outcomes of keeping a drain inside the wound postoperatively for 2-3 weeks for the treatment of lymphangioma in children.

Materials and method:

This prospective study was performed from July 2008 to June 2013. A total of 21 children were treated during the period. Complete surgical excisions were done in all patients and a drain tube was kept inside the wound (in situ) for 2 to 3 weeks. Among the cases 14 (66.6%) were male child and 7 (33.4%) were female, lowest age
operated was 1 month 15 days and highest age was 9 years. According to anatomical location, 7 of the lesion were in axillary region (33.33%), 5 (23.8%) were in neck region, 3 (14.28%) in chest wall, 3 (14.28%) in abdominal wall, 2 (9.52%) in upper limb and 1 (4.76%) in buttock.

Ultrasound were performed in all patients, CT scan in 5 patients and Duplex study in 3 patients only and histopathology were done in all patients.

Patients with intra-abdominal lymphangioma were not included in this study.

Result:

21 patients were operated during the study period and drain tube with 10fr-12fr feeding tube kept in (inside the wound) post operatively in all patients. Average duration of keeping drain tube was 18.57 days. Patients were released from hospital on 3rd-5th POD and asked to come for follow up on 7th, 14th and 21st POD and in between according to need of the patient. Parents were counseled to evacuate drainage bag every day, drain tube kept inside till no collection in drainage bag. Among the 21 cases 2 patients developed mild infection and 1 patient developed infection with pus formation and resolved after dressing and changing antibiotic. 1 patient developed recurrence (4.76%).

Discussion:

In this study we have clearly shown that keeping drain (drainage tube) in situ for 2-3 weeks is an effective, successful measure in addition to total surgical excision to prevent recurrence in the treatment of cystic hygroma. The exact mechanism by which it prevents recurrence is unknown but proven practically. As we have seen vacuum-assisted closure devices accelerate healing of many types of open wounds and minimize the adverse effects of secretion, Keeping drain tube in situ may have the similar effect.

Previously there was recurrence rate of about 50% and we used intralesional sclerotherapy in recurrence cases as it is difficult to motivate parents for re-operation. Our protocol was to keep drain tube in the wound cavity for a short (usually 3-5 days) period after operation. In one of our patient aged 3 years, having cystic hygroma in left upper extremity, patient discharged after operation keeping drain in situ and advised to come on 5th POD but patient failed to come on the day, instead came after 20 days and drain was inside the wound. Wound apparently looking healthy, no collection of fluid inside but on removal of drain tube small amount of purulent discharge was there, patient discontinued oral antibiotic and was afebrile. This patient then followed up closely with wound dressing and oral antibiotic for 7 days and there was no recurrence in 6 month period.
Because of incidental non recurrence of initial patient, we continued to apply this technique of keeping drain in situ along with oral antibiotic to our subsequent patients undergoing excision of complex lymphangioma.

This technique has been applied to cystic hygroma of all parts of body including neck, axilla, chest wall, abdominal wall and extremities. We keep patient admitted for 3-5 days after operation and then discharge with advice to come on 7th POD keeping drain tube in situ and advised to empty drainage bag daily, checking wound and advised to come on 14th POD for another check up. On 14th POD we inquire about the drainage, look for infection and decide whether drain tube should be kept or removed. If it is seen drainage continue to accumulate, drain kept for another week and patient is followed up on 21st POD. In our experience no drainage collection seen after 3 weeks, wound completely healed by this time. Patient is followed up 3 monthly up to 6 month and asked to contact if any complication or recurrence. There were negligible complications and recurrence rate was very low: only in 1 patient (4.76%), in comparison to the published data of recurrence rate of 10-40%9.

Recently used sclerotherapy OK-432 is very expensive and not easily available and has an approximately 60% response rate10. Intraliesional Bleomycin has a reported rate of complete resolution in only 50% of cases, and has a variety of complications including infection, gastrointestinal disturbances and pulmonary fibrosis11. Sclerotherapy with doxycycline needs close monitoring as patient may develop hypoglycemia with elevated systemic level of doxycycline and patient should be monitored for metabolic acidosis12,13.

We performed this small study, introduces several potential pitfalls. First, number of cases is small; 2ndly follow up of many of these patients is fairly short and irregular that possibility of late recurrences may be underestimated. Third, the exact mechanism of action is not yet to be established.

It can be presumed that longtime placement of drainage tube may minimize the adverse effects of secretion, initiate fibrosis and may induce low infection with low virulent streptococci and hence act like OK-432, which needs further study to comment finally.

Conclusion:

Despite the potential drawbacks, our success in the treatment warrants further verification of potential benefits of keeping drain tube in situ for 2-3 weeks after surgical excision of cystic hygroma.

References:


