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USE OF BLEOMYCIN IN CYSTIC HYGROMA: A SURGEON'S DILEMMA

Dental Science

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ABSTRACT

Cystic hygroma is a benign tumour of lymphatic origin that commonly occurs in the neck and presents mostly at birth. A variety of treatment options are available for the treatment of cystic hygroma. However, surgical enucleation of the lesion is the most suitable treatment option. Use of sclerosing agents such as intralesional bleomycin injections with some success rates have also been documented in literature. However, their use is also associated with difficulty in identifying the dissection planes intraoperatively.

KEYWORDS

cystic hygroma, lymphangioma, intralesional bleomycin

INTRODUCTION

Cystic hygroma is a congenital malformation of the lymphatic system that presents mostly at birth[1]. It is a benign tumor of lymphatic origin that occurs commonly in the neck and axilla. Majority of the cervical tumors are present in the posterior triangle of the neck usually beneath the platysma[2]. Lesions in the anterior triangle of neck may extend into the submandibular and submental spaces resulting in swelling in the floor of the mouth , which may or may not involve the tongue[3].

Cervical cystic hygromas may cause dysphagia and respiratory embarrassment , thus necessitating immediate surgical intervention[2].

Although surgical excision of the tumor is the treatment of choice for most clinicians, however, intralesional injections of bleomycin fat emulsion is another suitable alternative [4].

In this report, we are presenting two cases of cervical cystic hygroma both of which were treated surgically. However,the line of treatment was slightly different in one from the other as preoperatively bleomycin injections were administered in one of the two cases , in an attempt to reduce the size of lesion.

CASE REPORT 1

A 5 years old child reported to our hospital with the complaint of swelling and pain in right side of neck since birth. Patient was reportedly born with a small nodule in the right submandibular region that gradually increased in size alongwith development of pain in the same region.

Figure 1. Swelling in right submandibular region present since birth

On clinical examination, the swelling was soft, diffuse ,non tender, compressible and reducible in nature. A clinical diagnosis of cystic hygroma was made. CT scan was advised which showed a large cystic lesion with distinct boundaries. The patient was admitted and routine blood investigations were done, all of which were within normal limits. An excisional biopsy of the lesion under general anesthesia was planned . An incision was placed along the neck crease and soft tissue wall of the lesion was exposed. The lesion was carefully dissected from the posterior triangle of neck saving the spinal accessory and greater auricular nerve. It was removed from the undersurface of sternocleidomastoid muscle and carefully dissected around the internal jugular vein, common carotid artery, external carotid artery and submandibular region. Hemostasis was achieved using electrocautery and ligation. Closure of wound was obtained in layers. Postoperatively the child was kept under observation and care for a period of 2 days during which intravenous antibiotics and analgesics were administered. Her hospital stay was uneventful. Histopath ological reports were suggestive of cystic hygroma. Postoperatively, there was a gradual reduction in the size of the swelling and the child showed definite improvement. The patient is kept on regular follow up.

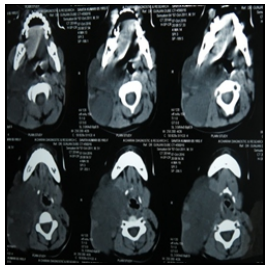

Figure 2. CT scan showing a cystic lesion in right cervical region.

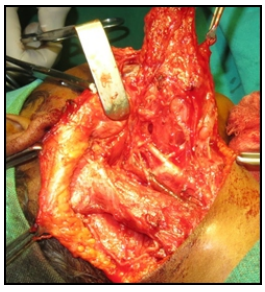

Figure 3. Intraoperative picture showing various anatomical structures after clearance of the cystic lesion

Figure 4. Excised specimen

Figure 5. Uneventful healing at surgical site with post surgical scar

CASE REPORT 2

Another child of 18 months age reported to our OPD with complaint of swelling in the neck since birth. The swelling was reportedly small in size initially and had progressed over a period of few months . There was no history of dyspnoea but the swelling was associated with difficulty in swallowing milk .

On clinical examination, there was a tense swelling involving the submandibular region bilaterally extending upto the floor of the mouth with gross elevation of the tongue which gave a clinical impression of cystic hygroma or a plunging ranula. Around 5ml of reddish , slightly turbid fluid was aspirated from the swelling and sent for biochemical analysis. The reports revealed a glucose content of 72 mg/dl and a protein content of 4.8gm/dl in the fluid which were not very diagnostic of any specific pathology. A CT scan of head and neck was done which showed a large expansile cystic lesion in right submandibular region with slight extension to the opposite side as well. Based on these findings, a clinical and radiographic diagnosis of cervical cystic hygroma was made. Intralesional bleomycin injections were administered to the child at regular intervals of 10 days and the child was kept on follow up. After a total of 4 intralesional bleomycin injections, a significant reduction in the size of the swelling was observed . Thereafter, a surgical excision of the lesion was planned under general anesthesia and the child was admitted in the hospital. Intraoperatively, the lesion was exposed through a neck crease incision. Skin flaps were raised and boundaries of the lesion were identified . However, dissection planes were not very distinct due to some amount of fibrosis induced in the subcutaneous tissues by administration of bleomycin intralesionally. The lesion was removed in toto and sent for histopathological examination , reports of which were suggestive of cystic hygroma. The child was kept under observation for a period of 7 days postoperatively and discharged thereafter. His hospital stay was uneventful.

Figure 6. Preoperative photograph showing swelling in right cervical region since birth.

Figure 7. Immediate preoperative photograph showing slight reduction in swelling following administration of bleomycin injections.

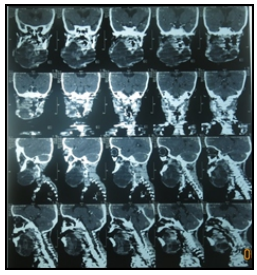

Figure 8. CT scan showing large cystic lesion in right submandi bular region.

Figure 9. Intraoperative photograph showing the incision markings

Figure 10. Intraoperative photograph showing the operative site after clearance of the lesion from the adjacent structures

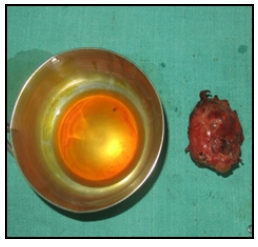

Figure 11. Resected specimen alongwith the cystic fluid content

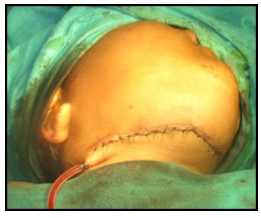

Figure 12. Closure of surgical site

Figure 13. follow up photograph showing healed surgical site

DISCUSSION

Cystic hygromas are rare benign lesions that present mostly at birth[1]. The most predominant site for their occurrence is the posterior triangle of neck owing to its extensive lymphatic system. The lesions may vary from 1-30 cm in size and may present as a painless enlarging mass that may increase in size thus causing dysphagia and respiratory embarrassment[2]. Surgical excision remains the treatment of choice for these lesions. However, aspiration may be done to decompress the lesion in those cases that compromise the airway[5]. Radiotherapy has proven to be ineffective in cases of cystic hygroma owing to the radioresistant nature of these lymphangiomas . Although radiotherapy was used in the past as a treatment modality , but its use has been abandoned nowadays due to the risk of malignant transformation associated with it[6]. Other alternate treatment modalities include use of sclerosing agents such as boiling water, sodium morrhuate, alcohol and 50% dextrose. However, these are associated with unpredictable results and may result in extensive sclerosis that may make future surgical intervention difficult. Nevertheless, sclerotherapy with bleomycin has been used with some success in cystic hygromas especially in children. These non surgical treatment options may be beneficial in children especially for treating lesions near parotid region in order to avoid the complications associated with surgery. Staged surgical excision can be done in some selected cases to avoid damage to certain vital anatomical structures.

CONCLUSION

Surgical enucleation is the treatment of choice for cystic hygroma. However, the proximity of the lesion to important anatomical landmarks makes dissection of the lesion difficult. Furthermore, the use of sclerosing agents such as bleomycin add to the level of difficulty encountered in identifying the planes of dissection intraoperatively. Hence, the risks of using sclerosing agents preoperatively should be weighed against the benefits associated with them.

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