CYSTIC HYGROMA: ANAESTHETIC CONSIDERATIONS AND REVIEW

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ABSTRACT

A 3-day-old child presented with a gross swelling on the right side of the neck extending beyond the midline and inability to swallow. It was diagnosed to be cystic hygroma and decision was made to excise the swelling as an emergency operation to enable the child to swallow and thrive better. During the gaseous induction, difficulty was encountered in maintaining the airway and subsequently to intubate the patient. Intraoperatively the surgeon was not able to excise the tumour completely. Postoperatively it was decided to ventilate the child electively because of the intubation difficulties encountered and not so firm floor of the mouth because of surgical excision. The stay in the neonatal ICU was marked with infection and facial nerve palsy.

Keywords: *Cystic hygroma, difficult intubation, surgical problems, postoperative management.*

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INTRODUCTION

Cystic hygromas in the cervical region present a challenge to the anaesthetist. Different techniques or "tricks" have been described to deal with tumours of head and neck which may present considerable difficulty with induction and maintenance of anaesthesia.

The literature does not abound in the description of management of extensive cervical cystic hygromas. Macdonald[1] reported two cases aged 2½ years and 22 years and discussed some of the problems of surgery and anaesthesia. There have been reports of cystic hygroma [2] and of pre-epiglottic cysts[2] causing respiratory obstruction in the neonate, but in only two reports[2,3] were involvement of hygroma in the epiglottis or larynx described. A case is reported of a neonate presenting with moderate-sized cystic hygroma along with the review of the features of cystic hygroma and anaesthetic management.

CASE REPORT

A baby girl weighing 3.45kg was delivered on 11th August 1991 by Caesarean section done for foetal distress. At birth a swelling was noticed on the right side of the neck which progressively increased in size. The tongue was pushed upwards and the child had difficulty in swallowing. For further management this child was transferred to the University Hospital.

Examination revealed swelling on both sides of the base of the tongue which pushed the tongue upwards. In the neck, the swelling was soft, 10cm x 6cm in size (Fig 1 and 2), and non tender. The skin over the swelling looked normal with

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Fig 1 – Photograph of the patient with cystic hygroma just before surgery

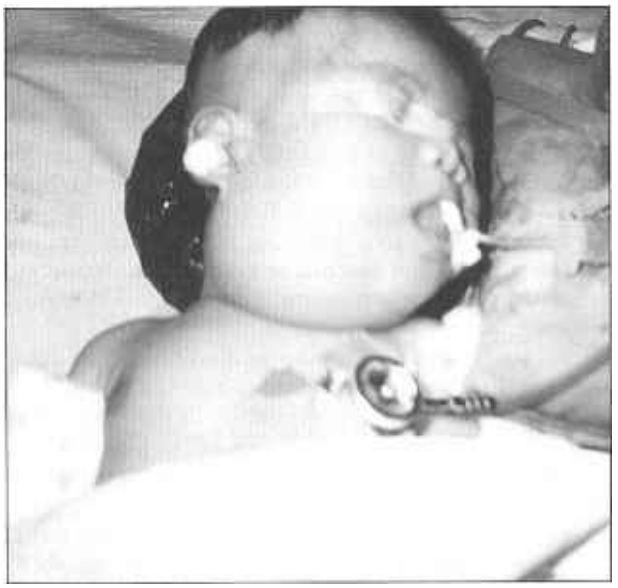
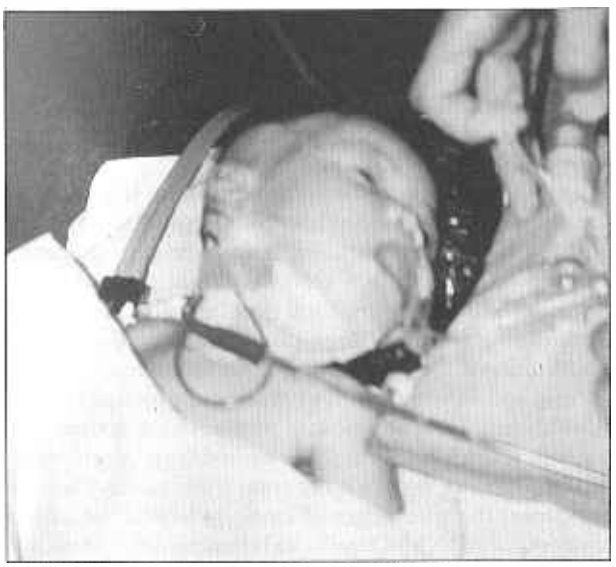


Fig 2 – Photograph showing the patient immediately after excision of cystic hygroma



no rise in temperature. The swelling was not attached to the skin, nor fixed to the deeper tissues. Transillumination was positive and there was a connection between the swelling in the neck and the tongue.

On 3rd September the patient developed severe inability to swallow and the surgeons decided to operate as an emergency case. The nasogastric tube inserted earlier was aspirated. Oxygen and halothane were administered by face mask. Laryngoscopy was then performed. It was difficult to push the tongue to the other side because of swelling under the base of the tongue. The larynx could not be visualised in the first attempt. The surgeon was asked to aspirate some fluid but only few millilitres could be aspirated. Laryngoscopy was tried again and this time the glottis could be seen and a 2.5 mm tube was introduced.

Anaesthesia was subsequently uneventful. Ventilation was controlled using 33% nitrous oxide in oxygen, norcuron and Jackson Rees modification of the Ayre's T piece. The cysts, which were multiloculated and extending to the floor of the mouth and to the tracheal wall were dissected and excised. However some of the cysts in the floor of the mouth could not be excised. Blood loss of about 150ml was replaced. At the end of the surgery norcuron was reversed with atropine 0.06mg and neostigmine 0.25mg. Though the patient was breathing well, extubation was deferred. During transfer from the operating theatre (OT) to the paediatric Intensive Care Unit (ICU) the patient extubated herself. After that she was put in a head box with oxygen under close observation. The child maintained the airway with good saturation for one whole day. On the next day, 4th September, she was given morphine after which respiration became shallow and she had to be intubated again and ventilated. On 7th September the child was again extubated after weaning from the ventilator.

Recovery in the ICU was also marked with infection which was treated by antibiotics.

After extubation slight deviation of the tongue was noted along with partial obstruction of the mouth by the tongue. On 14th September right facial nerve palsy was noted along with some mild inspiratory stridor.

The patient was then referred to the occupational therapist for oral stimulation. The exercises were taught to the mother. Feeding continued with Ryle's tube. Finally on 18th September the patient was discharged with a Ryle's tube. The parents were informed about possible recurrence and asked to bring the child for follow-up with the occupational therapist and the surgeon.

DISCUSSION

Cystic hygroma, also called cavernous lymphangioma, is a histologically benign congenital tumour of lymphatic origin[4].

Most commonly found in the neck alone, it also occurs in other sites corresponding to primitive lymph sac locations such as the axilla, mediastinum, groin and retroperitoneum. Endothelial membranes sprouting from embryonically sequestered lymph vessels form fibrillae that penetrate into surrounding normal tissues, canalise and produce large, multiloculated cysts filled with serous secretions[5,6].

Bill and Summer[7] in 1965 presented a unified concept considering lymphangiomas and cystic hygromas as variations of a single entity. The embryologic development of the lymphatic system is not completely understood and therefore, the true origin of cystic hygromas remains a mystery. Sabin[8,9], McClure[10], and Huntington[11] drew some meaningful conclusions regarding the development of the lymphatic anomalies. Now the controversy centres around the two theories, the "centrifugal theory" of Sabin and the "centripetal theory" of McClure and Huntington. Whether centrifugal or centripetal in development, most agree that lymphangiomas derive from a growth anomaly or arrest in the normal development, whereby the peripheral lymphatic vessels fail to flow into the jugular sacs, or the jugular sacs fail to reunite with the venous system.

Sometimes cases of true lymphangiomas appear to have an acquired aetiology. Kennedy[12] describes four types: traumatic, infectious, iatrogenic and tumour.

Kennedy[12] describes four types of lymphangiomas: superficial cutaneous lymphangioma; cavernous lymphangioma with two divisions, loose and compact; cystic hygroma or cystic lymphangioma; and diffuse systemic lymphangioma or lymphangioma haemangioma.

Cystic hygroma or cystic type of lymphangioma is a lesion developing from the lymphatic tissue in areas where expansion can occur and large multiloculated cystic spaces can develop which may contain serous fluid, sero-sanguinous fluid, or frank blood. Classically, it is a fluctuant lesion which transilluminates brightly.

The hygroma usually appears during the first year of life, but may be present at birth. The most prominent sign of cystic hygroma is the presence of a mass. The size of the mass usually determines whether there was a delay in seeking medical attention. Most masses noted at birth or shortly thereafter are very large cysts that can cause respiratory obstruction and certainly arouse parental curiosity and anxiety. Interference with normal breathing and swallowing are usually the second or third symptoms to appear. Respiratory difficulty can vary from mild stridor to cyanosis and complete obstruction. Larger cysts can cause tracheal compression if extension to the mediastinum occurs. Lesions extending into the floor of the mouth and tongue displace soft tissues posteriorly into the oropharnyx causing obstruction of both breathing and swallowing. Tracheostomy may be required to relieve respiratory obstruction. It is not uncommon for the larger hygromas to have lymphatic blebs within the mucous membrane at the base of the tongue and the supraglottic area. After surgical removal of the cervical lesion, it may be necessary to perform endoscopy and to open or remove these internal mucosal cystic structures. Marsupalisation or removal of the cysts with a carbon dioxide laser can be attempted if airway obstruction and feeding remain a problem following the removal of the cervical hygroma. Dysphagia is caused by the encroachment of the cysts around the hypopharynx and oesophagus that compress these structures. Dysphagia can persist after surgery, not only because of internal cystic structures but also because of surgical interference with the neural innervation to the muscle tissue of the hypopharnyx and upper cervical oesophagus.

Inflammation and infection of the cysts can occur at any time, usually following an upper respiratory tract infection. The cystic lesion becomes inflamed, probably increasing rapidly in size due to the increase in lymph response. If left untreated, an abscess, sepsis, or both can develop. Immediate treatment with antibiotics will usually prevent serious complication. Surgical excision of a hygroma should be delayed for at least three months if infection has occurred prior to surgery.

A sudden increase in the size of the cyst might be the result of an infection or spontaneous or traumatic

haemorrhage into the cystic structure. The cyst has blood vessels that are in the walls as well as traversing through and among the separate cystic spaces. Our patient presented with a swelling gradually increasing in size which ultimately had to be operated upon because of dysphagia.

Surgery remains the most acceptable mode of treatment. Few studies suggest that cystic hygroma has a high probability of spontaneous regression. Most surgeons suggest 18 months to two years as the optimum time for surgery. However delaying surgery in a hygroma that continues to grow will make future surgery difficult.

Alternative methods of treating cystic hygromas that are not routinely recommended include: aspiration, injection of sclerosing agents, diathermy and radiotherapy. Irradiation or radon seed implantation has been used successfully in primary treatment of cystic hygromas. Radiotherapy may have a place in those cases where complete excision is not possible and recurrent or persistent disease is still causing significant symptoms.

Anaesthetic management may present the following problems:

a) Haemorrhage

b) Intrathoracic extension – All cases must have chest X-ray to exclude the presence of intrathoracic lesions. Depending on its extent, such a lesion may be approached from the neck or through a proper thoracotomy. Even in the absence of respiratory distress, cough, tachypnoea, retractions or stridor[13], physical examination should be made for thoracic and oral extensions of the tumour. Should the tumour be found in the mediastinum, further delineation with fluoroscopy[14], angiography and CT scan[15] may aid in defining cardiopulmonary involvement and changes with respiration.

c) Extension into the mouth – If the tumour has interfered with swallowing, the child may be malnourished or dehydrated; intravenous therapy is then required before surgery. It may be impossible to visualise the larynx or to intubate blindly. Partial aspiration of the cyst might facilitate intubation but would render surgery more difficult. A surgeon should standby during the induction to do tracheostomy, if required. Our patient had this complication of the cyst extending to the floor of the mouth, thereby rendering intubation difficult. Partial aspiration was attempted but was unsuccessful.

d) Involvement of pretracheal region – The management of a tracheostomy in an infant is always difficult and is worse when an emergency tracheostomy has been made through a cystic hygroma. There is always considerable drainage of lymph and infection is inevitable.

e) Post-operative respiratory obstruction – The wound must be drained to prevent haematoma formation. If a haematoma occurs despite the drain, it should be aspirated or the stitches cut. Reactionary edema may develop in the first few hours post-operatively and necessitate tracheostomy. If the tongue has been chronically distorted by the tumour it may be very floppy after the operation and fall back very readily. Surgical damage to hypoglossal nerve increases the danger from the mobile tongue. If any difficulty is anticipated a long stitch should be inserted through the dorsum of the tongue at the end of operation.

CONCLUSION

Cystic hygroma can present many problems – dysphagia to the baby, anxiety to the parents, difficulties to the surgeon and a challenge to the anaesthetist during intra- and post-operative period. On encountering such a case, the attending doctors must be wary of these possible problems, anticipate the difficulties, and have a definite plan of action in mind before proceeding with further management.

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