

# Unusual Intraoperative Presentation of a Huge Cystic hygroma in a Neonate: An Anaesthetic challenge

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## Abstract

We discuss successful management of 15 days old neonate who underwent surgical excision of huge cystic hygroma located on right side of neck extending up to right shoulder, axilla and lateral chest wall. Just after sevoflurane induction and intubation patient started desaturating. We tried intubation thrice with 1 size larger ETT. Every time just after successful intubation, patient started desaturating. Lastly we planned to continue with IPPV. Throughout the procedure, oxygen saturation was fluctuating between 80-96% until large proportion of cystic hygroma had been excised. After 4hrs of uneventful surgery, patient was extubated in the operation theatre. Thereafter patient was shifted to paediatric postoperative care unit for observation and discharged on 20th postoperative day.

**Keywords:** Cystic hygroma, neonatal anaesthesia, neonatal airway, hypoxia

## Introduction

Cystic hygroma (CH) is a developmental malformation arising from the vascular lymphatic system. These are benign, painless, soft and compressible tumour, commonly located at cervico-facial regions and axilla. 40-50% lesions are identified in infants, but 90% are identified by the age of 2. 1-2 It results from obstruction between the lymphatic and venous pathways, commonly, in the fetal neck, which leads to lymph accumulation in the jugular lymphatic sacs in the nuchal region. Huge CH in neonates presents further challenge to the anaesthesiologist. Anaesthetic concern includes difficulty in visualizing the airway (due to extrinsic and intrinsic pressure caused by tumor extension into the mouth, thoracic extension, haemorrhage, infection, involvement of pretracheal region causing distortion and obstruction of the airway), bleeding, post operative respiratory obstruction and coexisting anomalies (Down's syndrome, Turner's syndrome

and congenital cardiac defects). 8 We present a case of cystic mass on right site of neck, shoulder, axilla and chest with potential respiratory compromise in a neonate.

## Case report

A 15 days old female child weighing 3 kgs was posted for excision of CH. She presented to the paediatric surgery OPD with a big lump in right side of neck, shoulder, axilla and chest since birth. She was born by caesarean section and had moderate respiratory distress. There was no associated pain, fever or increase in size of the lump. Her general condition was poor with 120/min heart rate and 55/min respiratory rate. Antero-posterior (AP) and lateral X-ray of neck showed minimal compression and deviation of trachea to left side. X-ray chest showed soft tissue shadow on right side of chest compressing the rib cage. MRI showed a huge lobulated smoothly circumscribed, septate, multilobular, cystic mass involving postero-

lateral aspect of neck, periclavicular region, superior aspect of shoulder, axilla and lateral thoracic wall on the right side consistent with a large lymphangioma with internal haemorrhage. (Fig1) Mass effect was seen in the form of right lateral thoracic wall compression by the mass (Fig 1). Her Hb was 14.4g/dl with 11400 /mm<sup>3</sup> total leucocytes count, 3.8 lacs/mm<sup>3</sup> platelet, 150 mEq/L Na<sup>+</sup>, 5 mEq/L K<sup>+</sup>, 2.0 sec bleeding time, 3.40 sec clotting time, 13.6 sec prothrombin time and INR 1.01. Parents were explained about recurrence and anticipated difficulties and informed high risk consent was taken. In operation room, 24G peripheral line was secured and electrocardiogram, non invasive blood pressure monitoring, pulse oximetry, temperature probe were attached. After premedication with intravenous atropine 0.12 mg, intravenous Ketamine 5 mg and preoxygenation with 100% O<sub>2</sub> for 5min, patient was induced with O<sub>2</sub> and 4-6% sevoflurane and successfully intubated with 2.5mm size oral uncuffed ET tube.

Immediately after intubation, patient started desaturating. Immediately the tube was taken out and patient was reintubated successfully with 3.0 mm size oral

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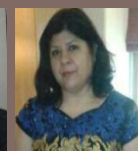
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uncuffed ET tube. Again the patient started desaturating and heart rate decreased to 65/min. Intravenous Atropine 0.12 mg was given. Again the tube was removed thinking of malpositioning, ventilated with facemask and reintubated by senior consultant with same size tube. After confirmation of proper placement, tube was secured. Again patient started desaturating. This time we continued IPPV for 5min. Dexamethasone was given to prevent supraglottic oedema. Saturation improved to 90%. Then we allowed the surgeon to start the surgery. Anaesthesia was maintained with O<sub>2</sub>-sevoflurane 2-2.5%. Patient's saturation was fluctuating between 80 - 96% till large proportion of CH had been excised. Then saturation improved to 100%. Further intraoperative course was uneventful. Surgery lasted for 4 hours. Patient was extubated on table and shifted to paediatric postoperative care unit. Postoperatively she was stable, conscious and crying. (Fig 2) She was discharged on 20th day.

### Discussion

CH is a benign tumor without potential malignancy with incidence of approximately 1/6000 live births. 70–80% of CHs occur in the neck, usually in the posterior cervical triangle. The remainder 20–30% occurs in the axilla, superior mediastinum, chest wall, mesentery, retro-peritoneal region, pelvis and lower limbs. 1, 3

The general time for surgery is known to be 18 months to 2 years of age. However, when the size of the lump is very large, a newborn infant may need surgery because it can cause respiratory distress or swallowing difficulty. Sometimes the size of even small

size CH can suddenly increase by infection, inflammation or internal hemorrhage and needed emergency surgery due to sudden respiratory arrest, hypovolaemic shock and sepsis. 1, 3, 4 In our case, the size was too large causing respiratory distress, thus needed emergency surgical excision.

Airway compromise due to CH, although rare, but remains an important cause of intraoperative mishap especially in an emergency situation.<sup>2</sup> For any patient needing removal of CH, it is most important for the anaesthesiologist to ruled out the extent of invasion by chest X-ray, CT scan and MRI and prepare accordingly for airway management. Besides this, intubation duration should be limited to < 20 seconds as neonates may rapidly move to a state of hypoxia when trying to intubate.<sup>1</sup>

Therefore, more preparation is required for infants with anatomical changes in the respiratory tract because of the cervical lump, but there are few methods to choose from.

Inhalation anaesthesia remains the preferred technique for management of a difficult pediatric airway.<sup>2</sup> We used it in our patient while maintaining spontaneous ventilation and patent airway reflexes. The neuromuscular blocking agents may relieve airway collapse during forceful expiration and straining but they may worsen the airway obstruction if the cause of obstruction is mechanical such as mediastinal mass. Therefore muscle relaxants were not used.

An initial drop in SpO<sub>2</sub> is sometimes seen immediately following intubation because children can take longer time to recover after lack of oxygenation during

laryngoscopy due to very low FRC and high O<sub>2</sub> consumption.<sup>5</sup> Sometimes large decrease in lung volume probably can lead to closure of small airways, development of intrapulmonary shunt and hypoxia. <sup>5</sup> So if all other indicators confirm endotracheal placement of ETT, the ETT should be left in place and ventilation continued for several breaths.<sup>6</sup> Though the exact mechanism of rapid development of hypoxia after intubation in our patient remained unclear but could be probably explained by large decrease in lung volumes due to compression of right side of chest by huge CH in already low FRC condition. The intraoperative accidental extubation or endobronchial intubation in case of extensive cervical manipulation of the newborn having CH has been reported extensively.<sup>1, 2</sup>

Extubation time is also a challenge as paralysis of the 7th, 11th and 12th cranial nerves was reported in 20% patients who underwent CH removal due to traction of the nerves during surgery and is generally reversible.<sup>1,3</sup> We had extubated our patient in the operation theatre because of adequate respiratory strength at the end of surgery.

### Conclusion

With proper preoperative evaluation and intraoperative management along with coordination and expertise of anaesthesiologist and paediatric surgeon, this complex situation can be managed successfully.

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