



ANESTHETIC MANAGEMENT OF IMPENDING AIRWAY COMPROMISE IN NEWBORN DUE TO CYSTIC HYGROMA

Anesthesiology

Dr. Kolhe S. D.*	Junior Resident, Dept. of Anaesthesia, Lokmanya Tilak Municipal Medical College & Hospital, Sion, Mumbai. *Corresponding Author
Dr. Desai U. H.	Associate Professor, Dept. of Anaesthesia, Lokmanya Tilak Municipal Medical College & Hospital, Sion, Mumbai.
Dr. Kadam R. R.	Junior Resident, Dept. of Anaesthesia, Lokmanya Tilak Municipal Medical College & Hospital, Sion, Mumbai.

ABSTRACT

A 6 days old female child presented with cystic lesion under surface of tongue on either side of frenulum with tongue pushed up with difficulty in breathing & feeding. It was diagnosed to be cystic hygroma and a decision was made to aspirate the content from the swelling to enable the child thrive better. GA with general endotracheal intubation planned after aspiration of cystic fluid. Around 13 cc fluid was aspirated with needle & syringe by paediatric surgeon which aided successful intubation in anticipated difficult airway. The intraoperative period was smooth. Uneventful extubation without any complications noted. Post operative recovery was smooth.

KEYWORDS

Cystic Hygroma, Difficult airway, Endotracheal intubation

INTRODUCTION

Hygroma means water-containing tumour in Greek. Cystic hygroma (CH), also known as lymphangioma, has its origins in the embryonic life, being an abnormality in the development of the lymphatic system.^(1,2,3) It represents 5% of the benign congenital malformations occurring during childhood.⁽⁴⁾ 50% of the cystic hygromas are present since birth and most of the remaining 50% appear by the age of 2-5 years.⁽⁵⁾ CHs are usually observed in children under two years old⁽⁶⁾; they are quite rare in adults and are thought to occur due to the proliferation of lymph vessels in response to trauma and/or head and neck infections.^(1,7,8)

Commonest site for cystic hygromas is the neck and tumor is the posterior triangle of the neck and it may involve vital structures, such as the sympathetic chain, carotid sheath content, and branches of the hypoglossal, lingual, and the facial nerves.⁽²⁾ other sites are axilla, superior mediastinum, retroperitoneum, mesentery pelvis, and lower limbs.⁽⁹⁾ Depending on anatomical site, they may cause dysphagia or airway obstruction and respiratory distress. Patient with cystic hygroma are frequently misdiagnosed as branchial cleft cysts. The lesion is the most often unilateral, and doughy on palpation. In children, cervical lymphangiomas are potentially life threatening due to direct localization or external compression on the airway.⁽¹⁰⁾

Significant differences exist between airways of the neonate and the adult. Anaesthetic management of the airway may be challenging in neonates and young infants with large neck mass like a huge cystic hygroma because these patients are at risk for sudden complete airway occlusion resulting in hypoventilation and hypoxemia. The aim of this report was to describe the difficulties encountered in intubation & anaesthetic management in newborn with cystic Hygroma.

Case Report:

A new born female of 6 days old of 6 days with 3 kg was diagnosed with cystic lesion in anterior neck of size 2.2*1.9*2 cm s/o cystic hygroma in Antenatal scan and presented with cystic lesion under surface of tongue on either side of frenulum with tongue pushed up.

Informant was the mother of the baby. She gave h/o that, baby was diagnosed with cystic lesion in anterior neck of size 2.2*1.9*2 cm s/o cystic hygroma in Antenatal scan. A baby born to P₂L₂, 22 year old mother. The baby was the second child of healthy unrelated parents; the medical history of the mother was unremarkable. Mother delivered baby by caesarean section due to severe oligohydrominos which was 3 kg full term female child at our Tertiary care center with swelling on under surface of tongue. Swelling lead to airway obstruction, it was decide to operate the child.

On General examination, the baby was moderate conscious & afebrile. Pulse rate 154 bpm, all peripheral pulses well felt, RR 65-70/ min, with sternal & subcostal retractions with respiratory distress with SPO₂ at

room air was 93-95%. On room air had noisy breathing in supine position and feel comparatively comfortable in lateral & prone position. Baby receiving formula milk via infant feeding tube.

On airway examination, the swelling was present under surface of tongue on either side of frenulum with tongue pushed up & no other significant craniofacial deformity found. (Figure 1). The swelling was soft on palpation, with poorly defined margins, nontender and when subjected to light test was brilliantly translucent.

On USG a large ill-defined superficial multicystic and multiseptate soft tissue lesion of size 3.4*2.5*4.3 cm was present in floor of mouth. It is predominantly cystic s/o lymphatic malformation of bae of tongue ?cystic hygroma. ON CECT of neck, an approx. 3.6*2.5*3.0 cm sized well defined non enhancing cystic lesion in floor of mouth in sublingual space abutting the geniohyoid muscle inferiorly causing mass effect on anterior belly of both right and left diaphragic muscle. The lesion is causing near complete obliteration of oral cavity.

Two thin septae noted in the anterior and inferior aspect of lesion. these features are likely s/o benign etiology like lymphatic malformation involving floor of mouth in sublingual space. Blood investigation within normal limit.

Pre-operative evaluation was thoroughly carried out after taking informed consent from mother of newborn. Then newborn taken to the operation theatre where standard monitors were attached and supplemental oxygen was administered via nasal canula at 2L/min, Intravenous line secured.

A rescue tracheostomy by surgeon was available as a standby during induction. Baby was kept NBM. Because cystic hygroma present with difficult airway challenge to the anaesthesiologist, a difficult airway cart was kept ready. Around 13 cc fluid was aspirated with needle & syringe by paediatric surgeon under local anaesthesia..

The child was premedicated with intravenous atropine 0.01 mg/kg. A shoulder roll was used to keep the child at optimal laryngoscopic position. The child was induced with oxygen, sevoflurane, inj propofol 1 mg/kg & inj atracurium 0.75 mg/kg.

The general anaesthesia maintained with Oxygen, air, sevoflurane & intermittent atracurium with JR circuit with controlled manual ventilation. Intra-operatively child was stable.

At the end of surgery after spontaneous attempt of respiration, the child was reversed with inj Neostigmine 0.05 mg/kg & Atropine 0.02 mg/kg.

The child was shifted to postoperative recovery room for observation and further monitoring after return of adequate muscle power, respiratory efforts, cry and movements.



Fig 1 : Swelling undersurface of tongue S/O Cystic Hygroma



Fig 2 : Airway Examination



Fig 3: Aspiration of fluid from Cystic Hygroma



Fig 4 : Picture after aspiration of cystic fluid

DISCUSSION:

This report describes a rare case of a newborn female presenting with a neonatal Cystic Hygroma underneath surface of tongue, which impeded breathing & breast-feeding.

Around 90 cases have been reported in the literature, it is difficult to determine the incidence of these lesions because of varied nomenclature, such as "anterior tongue median cyst,"⁽¹¹⁾ "bronchogenic cyst,"⁽¹²⁾ "gastric heterotopias,"⁽¹³⁾ "enterocystoma,"⁽¹⁴⁾ and "heterotopic gastrointestinal cyst."⁽¹⁵⁾ In this report, the authors preferred to use the "Cystic Hygroma. Hs are benign congenital malformations of the lymphatic system that manifest as macrocystic simple or multiple tumors, having a low communication with normal lymphatic vessels or venous structures. A very important percentage (about 90%) of hygromas is diagnosed in children less than two years old."^(16,17)

As stated in many studies, most of the CHs develop in the cervical and facial regions (70–75%), axilla, mediastinum, groin, and more rarely

in the liver, spleen, kidneys, intestines, mesentery, caul, adrenal glands, thoracic wall, limbs, etc.^(18,19). Until now, the exact pathogenesis of malformations in the lymphatic system is not completely known. There were proposed two hypotheses regarding the genesis of lymphatic malformations: an abnormal or insufficient network of the lymphatic system or a deficient connection between the lymphatic system and the venous system^(20,21). The hypothesis of a deficient connection between the lymphatic system and the venous system is more probable for CHs of the head and neck, as the two vascular systems present some common stages and connections during the embryo development.⁽²²⁾

Cystic hygromas are benign lesions & if asymptomatic, they do not necessitate treatment. In indications for treatment include infection, haemorrhage, respiratory distress, dysphagia or disfigurement. The mainstay of management is surgical. At present, there are multiple treatment options: conservatory treatment by injecting sclerosing agents (Bleomycin, Rifampicin, corticosteroids, Dextrose, etc.), repeated content aspiration, radiotherapy, ablation by radiofrequency, surgical removal, etc.^(23,24). Treatment options will be individualized depending on the size, anatomical location & complications of the lesion.

Cystic hygroma present several challenges regarding airway management. This case highlights the importance of functional complications of cystic hygroma that must be considered for peri-operative airway management. In our case as this benign lesion was obliterating oral cavity, to secure the airway needle decompression of large cystic components was planned to decrease the risk of cyst rupture before attempting to secure the airway and it turned out to be successful and valuable additions to management. It made difficult mask ventilation & intubation smooth & easier. To prevent aspiration & bronchospasm, the cyst was aspirated under local anaesthesia. If cyst was non aspirable, the spontaneous induction would have been done with inhalational agent after check scopy and muscle relaxant given only after intubation. Plan C included awake fiberoptic intubation with appropriate size fiberoptic scope. The results were favourable, as there were not recorded any immediate complications and the patient's monitoring for a period of one years showed no presence of local relapse.

CONCLUSION:

In our report, we presented a case of Sublingual CH in a 6 days old girl child, investigated by USG & CECT, which had challenging airway management which was managed with method of aspiration of cyst content to make intubating conditions favourable. The objective in surgery of CH was preventing the obstruction of breathing & difficulty for feeding of child. The absence of immediate complications and local recurrence for one years after surgery show that we chose the best treatment method.

REFERENCES:

1. Aneeshkumar MK, Kale S, Kabbani M, David VC. Cystic lymphangioma in adults: can trauma be the trigger? *Eur Arch Otorhinolaryngol*, 2005, 262(4):335–337. <https://doi.org/10.1007/s00405-004-0780-6> PMID: 15841412
2. Guruprasad Y, Chauhan DS. Cervical cystic hygroma. *J Maxillofac Oral Surg*, 2012, 11(3):333–336. <https://doi.org/10.1007/s12663-010-0149-x> PMID: 23997487 PMID: PMC3428451
3. Eivazi B, Werner JA. Extrakranielle vaskuläre Fehlbildungen (Hämangiome und vaskuläre Malformationen) im Kindes- und Jugendalter – Diagnostik, Klinik und Therapie [Extracranial vascular anomalies (hemangiomas and vascular malformations) in children and adolescents – diagnosis, clinic, and therapy]. *Laryngorhinotologie*, 2014, 93(Suppl 1):S185–S202. <https://doi.org/10.1055/s-0033-1363216> PMID: 24710783
4. Gaddikeri S, Vattoth S, Gaddikeri RS, Stuart R, Harrison K, Young D, Bhargava P. Congenital cystic neck masses: embryology and imaging appearances, with clinicopathological correlation. *Curr Probl Diagn Radiol*, 2014, 43(2):55–67. <https://doi.org/10.1067/j.cpradiol.2013.12.001> PMID: 24629659
5. Emery PJ, Bailey CM, Evans J. Cystic hygroma of the head and neck. A review of 37 cases. *J Laryngol Otol*. 1984;98(6):613–9. doi:10.1017/s0022215100147176.
6. Derin S, Şahan M, Dere Y, Çullu N, Şahan L. Cervical cystic hygroma in an adult. *Case Rep Pathol*, 2014, 2014:209427. <https://doi.org/10.1155/2014/209427> PMID: 25548704 PMID: PMC4273562
7. Sherman BE, Kendall K. A unique case of the rapid onset of a large cystic hygroma in the adult. *Am J Otolaryngol*, 2001, 22(3):206–210. <https://doi.org/10.1053/ajot.2001.23430> PMID: 11351291
8. Kasapoğlu F, Yildirim N. Cystic hygroma colli in adults: a report of two cases, one with atypical location. *Kulak Burun Bogaz İhtis Derg*, 2008, 18(5):326–329. PMID: 19155682
9. Pandit SK, Rattan KN, Budhiraja S, Solanki RS. Cystic lymphangioma with special reference to rare sites. *Indian J Pediatr*. 2000;67(5):339–41. doi:10.1007/BF02820682
10. Thompson DM, Kasperbauer JL. Congenital cystic hygroma involving the larynx presenting as an airway emergency. *J Natl Med Assoc*, 1994, 86(8):629–632. PMID: 7932844 PMID: PMC2607717
11. Lister J, Zachary RB. Cystic duplications in the tongue. *J Pediatr Surg* 1968;3:491Y493
12. Boue[†] DR, Smith GA, Krous HF. Lingual bronchogenic cyst in a child: an unusual site of presentation. *Pediatr Pathol* 1994;14:201Y205

13. Johnston C, Benjamin B, Harrison H, et al. Gastric heterotopia causing airway obstruction. *Int J Pediatr Otorhinolaryngol* 1989;18:67Y72
14. Grime PD. Giant enterocystoma within an infant's tongue. *J Laryngol Otol* 1990;104:814Y818
15. Martone CH, Wolf SM, Wesley RK. Heterotopic gastrointestinal cyst of the oral cavity. *J Oral Maxillofac Surg* 1992;50:1340Y1342.
16. Manikoth P, Mangalore GP, Megha V. Axillary cystic hygroma. *J Postgrad Med*, 2004, 50(3):215–216. PMID: 15377810
17. Lerat J, Mounayer C, Scomparin A, Orsel S, Bessede JP, Aubry K. Head and neck lymphatic malformation and treatment: clinical study of 23 cases. *Eur Ann Otorhinolaryngol Head Neck Dis*, 2016, 133(6):393–396. <https://doi.org/10.1016/j.anorl.2016.07.004> PMID: 27497629
18. Kocher HM, Vijaykumar T, Koti RS, Bapat RD. Lymphangioma of the chest wall. *J Postgrad Med*, 1995, 41(3):89–90. PMID: 10707725
19. Kaur N, Gupta A, Amratash, Singh N. Giant cystic hygroma of the neck with spontaneous rupture. *J Indian Assoc Pediatr Surg*, 2007, 12(3):154–155. <https://doi.org/10.4103/0971-9261.34959> <https://www.jiaps.com/text.asp?2007/12/3/154/34959>
20. Dhrif AS, El Euch D, Daghfous M, Cherif F, Mokni M, Dhahri ABO. Malformation lymphatique macrokystique (lymphangiome kystique) du membre supérieur : à propos d'un cas [Macrocystic lymphatic lymphangioma (cystic lymphangioma) of the upper extremity: a case report]. *Arch Pediatr*, 2008, 15(9):1416–1419. <https://doi.org/10.1016/j.arcped.2008.06.003> PMID: 18667296
21. Wiegand S, Eivazi B, Barth PJ, von Rautenfeld DB, Folz BJ, Mandic R, Werner JA. Pathogenesis of lymphangiomas. *Virchows Arch*, 2008, 453(1):1–8. <https://doi.org/10.1007/s00428-008-0611-z> PMID: 18500536
22. Eivazi B, Werner JA. Extracranial vascular malformations (hemangiomas and vascular malformations) in children and adolescents – diagnosis, clinic, and therapy. *GMS Curr Top Otorhinolaryngol Head Neck Surg*, 2014, 13:Doc02. <https://doi.org/10.3205/cto000105> PMID: 25587362 PMCID: PMC4273163
23. Kamath BS, Chatterjee AS, Chandorkar I, Bhanushali H. Giant recurrent cystic hygroma: a case report. *J West Afr Coll Surg*, 2014, 4(2):100–111. PMID: 26587526 PMCID: PMC4500765
24. Janardhan HP, Saheera S, Jung R, Trivedi CM. Vascular and lymphatic malformations: perspectives from human and vertebrate studies. *Circ Res*, 2021, 129(1):131–135. <https://doi.org/10.1161/CIRCRESAHA.121.319587> PMID: 34166069 PMCID: PMC8238353