Difficult Airway Management in Pediatric with a Large Cystic Hygroma Colli Undergoing One-Stage Excision Surgery

Mahendratama Purnama Adhi¹, Hery Poewrosusanta²
¹Department of Anesthesiology and Intensive Care, Faculty of Medicine, Lambung Mangkurat University/Ulin Hospital, Banjarmasin, Indonesia
²Pediatric Surgery Consultant, Department of Surgery, Faculty of Medicine, Lambung Mangkurat University/Ulin Hospital, Banjarmasin, Indonesia

ABSTRACT

Background: A large cystic hygroma colli is a complicating factor in airway management in pediatric. Proper preparation, planning, and anticipation can reduce the risk of complications, morbidity, and mortality during difficult airway management. The fundamental principle of difficult airway management is maintaining adequate oxygenation and avoiding hypoxemia by maintaining spontaneous ventilation.

Case: A 22-month-old girl, weighing 9 kg, diagnosed with a large cystic hygroma colli who underwent single-stage excision surgery. Cystic hygroma was experienced since birth, and with age, the cysts enlarge to a size of 25 x 17 x 12 cm, extending towards the face and shoulders. The patient did not experience stridor and symptoms of airway obstruction. The chest x-ray revealed no cyst extension into the chest cavity and showed minimal tracheal deviation to the right. The difficult airway management was accomplished while maintaining the patient's spontaneous breathing. Sedation and analgesia were obtained with intravenous administration of dexmedetomidine, ketamine, and nebulized lidocaine. A video laryngoscope is used to facilitate intubation. Awake extubation was performed after a cuff-leak test confirmed that there was no risk of laryngeal edema, laryngeal nerve injury, or tracheomalacia.

Conclusion: A difficult airway in a pediatric patient with a large cystic hygroma colli can be managed successfully by maintaining spontaneous breathing, achieving optimal levels of sedation and analgesia with dexmedetomidine, ketamine, and nebulized lidocaine, and the use of a video laryngoscope.

Keywords: cystic hygroma colli, dexmedetomidine, difficult airway management, ketamine, pediatric

INTRODUCTION

The presence of complicating factors in pediatric airway management is associated with increased morbidity and mortality. Early recognition of risk factors and anticipation of difficult airways, and good preparation and planning can reduce the risk of complications during difficult airway management in pediatrics.¹

Cystic hygroma or lymphangioma cyst is a benign congenital malformation of the lymphatic system due to obstruction between the lymphatic pathways and veins, causing lymph accumulation in the jugular lymphatic sac in the nuchal.² The incidence of cystic hygroma is 1: 200,000 births, and over 50% of cystic hygroma is seen at birth, and 80-90% occur at two years of age. A large cystic hygroma and expansion into the mediastinum can cause acute airway.³

Pediatrics with a large cystic hygroma is one of the predictive factors for difficulty in airway management during anesthesia. This presents a challenge for anesthesiologists. The key to successfully managing a difficult airway is inseparable from identifying potential problems and considering several airway managements.² Here, we describe airway management in a pediatric with a large cystic hygroma colli who underwent one-stage excision surgery.

Case Report: A 22-month-old girl, weighing 9 kg, diagnosed with a large cystic hygroma colli who underwent single-stage excision surgery. Cystic hygroma was experienced since birth, and with age, the cysts enlarge to a size of 25 x 17 x 12 cm, extending towards the face and shoulders. The patient did not experience stridor and symptoms of airway obstruction. The chest x-ray revealed no cyst extension into the chest cavity and showed minimal tracheal deviation to the right. The difficult airway management was accomplished while maintaining the patient's spontaneous breathing. Sedation and analgesia were obtained with intravenous administration of dexmedetomidine, ketamine, and nebulized lidocaine. A video laryngoscope is used to facilitate intubation. Awake extubation was performed after a cuff-leak test confirmed that there was no risk of laryngeal edema, laryngeal nerve injury, or tracheomalacia.
CASE

We have obtained written consent from the patient’s parents to include in this report. A 22-month-old girl, weighing 9 kg, diagnosed with a large cystic hygroma colli who underwent one-stage excision surgery. According to both parents’ histories, the cyst in the patient’s neck has been present since birth and had become more prominent as the patient grew old. From the history of labor, the patient was born with the normal delivery, term, birth weight 3000 grams, cried immediately, and had no history of cyanosis or congenital heart defects. Physical examination revealed a cyst mass on the right neck measuring 25x17x12 cm, extending towards the face and shoulders (Figure 1).

![Figure 1](image1.png) **Figure 1.** A. Anterior view of cystic hygroma colli. B. Lateral view of cystic hygroma colli

There was no stridor or other signs of airway obstruction. Her pulse rate was 112 beats per minute, her blood pressure was 80/50 mmHg, her respiratory rate was 28 times per minute, and SpO$_2$ was 98% in room air. There were no heart defects or any congenital abnormalities discovered. The laboratory results obtained blood hemoglobin level of 9.9 g/dL, hematocrit 34.8%, leukocyte count of 10,500/µl, a platelet count of 470,000/µl, and the results of blood coagulation, liver function, renal function, and electrolytes within normal limits. The chest x-ray reveals no cyst expansion into the thoracic cavity, and the trachea shifted to the right (Figure 2). The patient’s parents received an explanation of the risk of difficulty in airway management during anesthesia, the plan for airway management to be carried out, and the possibility of delaying surgery in the event of failure of airway management.

The patient was accompanied by her parents, and in the premedication room, she was given intravenous midazolam 0.05 mg/kg. Standard routine monitors (pulse, blood pressure, SpO$_2$, body temperature) are installed. Atropine sulfate 0.1 mg was given intravenously, followed by dexmedetomidine (loading dose 1 µg/kg in 10 minutes and followed by continuous infusion of 0.5 µg/kg/hour). During the loading dose of dexmedetomidine, 1% lidocaine nebulization was administered. After the patient was sedated (Ramsay score 4), we transferred to the operating room. Preoxygenation with 100% O$_2$ was given for 3 minutes using Jackson Reese at a flow of 6 L/min. Ketamine 0.5 mg/kg was given prior to the laryngoscopy procedure using a McGrath video laryngoscope (MAC VL, Medtronic) with an angulated blade number 2. The epiglottis can be viewed as a Grade 2 Cormack-Lehane by shifting the trachea to the left and applying some pressure (Figure 3). Intubation was performed using a 3.5 ID micro cuff endotracheal tube (ETT). After intubation, we placed additional intravenous access using a large-bore iv catheter.

Anesthesia was maintained with a continuous infusion of 0.5 µg/kg/h dexmedetomidine, 1.5 vol% sevoflurane in a mixture of 40% oxygen and 60% N$_2$O with spontaneous ventilation using Jackson Reese. There was bleeding of 110 ml during the surgery, and blood replacement was done with a packed red cell (PRC). When surgical wound closure was performed, continuous infusion of dexmedetomidine and N$_2$O was discontinued. Sevoflurane was stopped when the surgery was completed. We performed awake extubation after confirming no signs of laryngeal edema, laryngeal nerve injury, and tracheomalacia with a cuff-leak test. The cyst excision surgery and external drain placement lasted for 150 minutes and were performed uneventfully. We found no complications of postoperative facial and hypoglossal nerve injury. Using the FLACC scale (Face, Legs, Activity, Cry, Consolability), postoperative pain management with paracetamol 125 mg
intravenously every 6 hours resulted in a pain score of 2. The patient was admitted to the ICU for one day and underwent outpatient care on day 5.

**DISCUSSION**

In 25-75% of cases, genetic disorders often accompanied cystic hygroma. Cystic hygroma can be found in Turner’s syndrome, Down’s syndrome, Klinefelter’s syndrome, trisomy 18, and trisomy 13. The history and preoperative physical examination should focus on signs and symptoms of compressed airways, respiratory distress, cough, stridor, tachypnea, dysphagia, and dyspnea. In pediatrics undergoing cyst excision surgery, the anesthesiologist should be aware of the extension of the cyst into the respiratory tract and prepare a plan for managing the difficult airway. The enlargement of the cyst could push the tongue, and the presence of tracheal deviation can complicate visualization during direct laryngoscopy. The x-ray imaging, CT scan, and MRI of the chest and neck can assess the cyst’s size, position, extent, and tracheal deviation.

Securing the airway, keeping adequate oxygenation, and preventing hypoxemia were essential factors in this case. The principle is to keep the patient breathing spontaneously to avoid inability or failure of ventilation following multiple intubation attempts. In addition, maintaining spontaneous ventilation can awaken the patient quickly if necessary. Spontaneous ventilation can also show the position of the glottis through secretion bubbles that are visible on expiration.

With these considerations, we planned for intubation to be performed with a sedated patient, maintaining spontaneous breathing. Administering oxygen through the facemask using a Jackson Reese, and the use of a McGrath video laryngoscope to facilitate intubation.

Awake intubation in pediatrics can cause pain and discomfort so that the child becomes uncooperative and can cause psychological trauma. Providing sedation and analgesics can facilitate intubation and reduce the risk of injury. To address the patient’s anxiety, we provide intravenous midazolam when the child arrived at the premedication room. We use a combination of intravenous dexmedetomidine, ketamine, and lidocaine nebulization as sedation and analgesia to facilitate intubation. Dexmedetomidine is an α2-adrenergoreceptor agonist which has sedating, anxiolytic, and analgesic effects. In contrast to other sedation regimens, the mechanism of action of dexmedetomidine in the locus coreolus of the central nervous system is similar to that of natural sleep without causing respiratory depression. Its ability to maintain airway tone and spontaneous breathing make dexmedetomidine an option for diagnostic procedures requiring sedation. In addition, dexmedetomidine can also blunt airway reflexes and lessen laryngospasm response, making it a sedative option for laryngoscopy and intubation in pediatric with predicted difficult airway management. In removing the corpus alienum using fiberoptic bronchoscopy under intravenous anesthesia with spontaneous breathing, the combination of dexmedetomidine and propofol showed a more stable hemodynamic and respiratory profile than the combination of remifentanil and propofol. In this case, we used ketamine as an adjunct to analgesia and sedation for intubation. The combination of dexmedetomidine and ketamine has shown optimal results for intubation in pediatric with Pierre Robin syndrome with Tetralogy Fallot and Treacher Collins syndrome. We chose ketamine because it has an analgesic effect without causing respiratory depression to maintain spontaneous breathing. The sympathomimetic nature of ketamine can balance out the bradycardia and hypotensive effects of dexmedetomidine. The dose of ketamine administration may also be lower due to the synergistic effect of dexmedetomidine. Administration of atropine sulfate can prevent hypersalivation due to ketamine, and other than that, dexmedetomidine can reduce the production of the salivary glands. Topical anesthesia by spraying it over the glottis can minimize airway reactivity during laryngoscopy and intubation. A topical anesthesia spraying can be done after a deep level of anesthesia has been reached. The application of topical anesthetics can also be made with a nebulizer. In addition, lidocaine nebulization can reduce the incidence of emergence agitation due to sevoflurane. The same effect is also owned by dexmedetomidine.

Using a video laryngoscope in pediatrics with difficult airway prediction can improve glottic visualization, the degree of Cormack-Lehane, and the success of intubation. However, the success rate and the required intubation time did not differ between the use of a video laryngoscope and a conventional Macintosh laryngoscope in pediatric patients without difficult airway management.

The ETT must be appropriately placed and fixed due to the potential of inadvertent release or extubation as a result of manipulation during surgery. Exubation in a patient with a cystic hygroma colli requires attention to several factors. Postoperative complications such as respiratory obstruction due to airway edema, tracheomalacia, edema of the tongue, and laryngeal nerve injury may occur. Exubation should be delayed in conditions where this risk of complications is identified. In our case, we performed extubation after the cuff-leak test confirmed that there were no probable complications of airway obstruction due to edema, tracheomalacia, or laryngeal nerve injury in our patient. We performed extubation when the child is fully awake and had active moving. Awake extubation can minimize the risk of laryngeal spasms. Close monitoring after extubation is still needed because postoperative laryngeal edema can occur because of obstruction of lymphatic flow that triggers an inflammatory response.

**CONCLUSION**

The presence of a large hygroma colli cyst can cause difficulties with airway management in pediatrics. Securing the airway, maintaining adequate oxygenation, and avoiding hypoxemia by maintaining spontaneous breathing are key principles in difficult airway management. Administration of dexmedetomidine, ketamine and nebulized lidocaine can provide adequate sedation and analgesia without causing respiratory depression. The use of a video laryngoscope improves epiglottic visualization and successful intubation. Extubation should be carried out carefully, and close monitoring after extubation is necessary because of the risk of postoperative airway obstruction.

**ACKNOWLEDGMENT**

-  

**CONFLICT OF INTEREST**
REFERENCES
1. Huang AS, Hajduk J, Rim C, Coffield S, Jagannathan N. Focused review on management of the difficult paediatric airway. *Indian J Anaesth*. 2019;63(6):428-436. doi:10.4103/ija.UA_250_19
2. Rao KS, Shenoy T. Anesthetic management of a large cystic hygroma in a newborn. *Anesth essays Res*. 2015;9(2):270-272. doi:10.4103/0259-1162.156364
3. Lei SY. Cystic hygroma. In: Houck PJ, Hache M, Sun LS, eds. *Handbook of Pediatric Anesthesia*. McGraw-Hill Education; 2015.
4. Gurulingappa, Awati MN, Aleem MA. Cystic hygroma: A difficult airway and its anaesthetic implications. *Indian J Anaesth*. 2011;55(6):624-626. doi:10.4103/0019-5049.90626
5. Fiadjoj E, Stricker PA, Litman RS. Pediatric Airway Management. In: Gregory GA, Andropoulos DB, eds. *Gregory's Pediatric Anesthesia*. Wiley-Blackwell; 2012.
6. Dilek O, Yasemin G, Atci M. Preliminary experience with dexmedetomidine in neonatal anesthesia. *J Anaesthesiol Clin Pharmacol*. 2011;27(1):17-22.
7. Mahmoud M, Mason KP. Dexmedetomidine: review, update, and future considerations of paediatric perioperative and periprocedural applications and limitations. *Br J Anaesth*. 2015;115(2):171-182. doi:10.1093/bja/avv226
8. Chen K-Z, Ye M, Hu C-B, Shen X. Dexmedetomidine vs remifentanil intravenous anaesthesia and spontaneous ventilation for airway foreign body removal in children. *Br J Anaesth*. 2014;112(5):892-897. doi:10.1093/bja/aet490
9. Dsouza S, Barad D, Barme S. Intubation using dexmedetomidine and ketamine in a child with Pierre Robin sequence. *J Anaesthesiol Clin Pharmacol*. 2012;28(4):545-546. doi:10.4103/0970-9185.101964
10. Irvani M, Wald SH. Dexmedetomidine and ketamine for fiberoptic intubation in a child with severe mandibular hypoplasia. *J Clin Anesth*. 2008;20(6):455-457. doi:10.1016/j.jclinane.2008.03.012
11. Hasanein R, El-Sayed W. The effect of nebulized lidocaine hydrochloride on emergence from sevoflurane anesthesia in children undergoing Tonsillectomy. *Egypt J Anaesth*. 2013;29(4):351-356. doi:https://doi.org/10.1016/j.ejga.2013.06.001
12. Dadure C, Sabourdin N, Veyckemans F, et al. Management of the child’s airway under anaesthesia: The French guidelines. *Anaesthesia, Crit care pain Med*. 2019;38(6):681-693. doi:10.1016/j.accpm.2019.02.004
13. Wallis C, Alexopoulou E, Antón-Pacheco JL, et al. ERS Statement on Tracheomalacia and Bronchomalacia in Children. *Eur Respir J*. Published online January 1, 2019;1900382. doi:10.1183/13993003.00382-2019
14. Kuriyama A, Jackson JL, Kamei J. Performance of the cuff leak test in adults in predicting post-extubation airway complications: a systematic review and meta-analysis. *Crit Care*. 2020;24(1):640. doi:10.1186/s13054-020-03358-8
15. Wijaya Ramlan A, Ang M, Peddyandhari F. Airway obstruction after anesthesia in a 3-month-old baby with lymphangioma. *Bali J Anesthesiol*. 2020;4(2):81-83. doi:10.4103/BJOA.BJOA_31_20