CASE REPORT

An Innovative Airway Management of an Anticipated Difficult Airway in a Pediatric Patient with Cervical Cystic Hygroma: A Case Report

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ABSTRACT

Airway management of a pediatric patient with cervical cystic hygroma can be challenging as the large neck mass may extend in the oral cavity, result in tracheal deviation, and cause possible upper airway obstruction. This is a case of a 4-year-old female admitted due to sudden enlargement of neck mass associated with dysphagia and sialorrhea. Patient was intubated under sedation while maintaining spontaneous ventilation with a pediatric flexible fiberoptic scope through a nasopharyngeal airway serving as a passage guide for ease of scoping and a protective device against trauma.

Keywords: cystic hygroma, airway management, intubation

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INTRODUCTION

Cystic hygroma is a benign cyst that develop due to congenital malformation of the lymphatic drainage system that typically form in the neck, clavicle, and axillary regions.[1,2] Although the exact embryonic origin is unclear, it is widely accepted to arise from remnants of embryonic lymphatic tissue which retains the potential for propagation and is capable of transgressing anatomical boundaries.[3]

It represents 6% of all benign lesions of infancy and childhood of which about 50% are reported present since birth and most of the remaining 50% appear by two years of age.[4] It manifests as large, deep, diffuse swelling that is doughy upon palpation and is trans-illuminant. It may grow rapidly due to accumulation of lymph itself, blood secondary to hemorrhage, or pus secondary to infection which can further cause dysphagia or airway obstruction and respiratory distress that requires immediate surgical intervention.[5]

If symptomatic, the mainstay of treatment is surgical. However, other treatment options include sclerotherapy, drainage, radiofrequency ablation or cauterization.[1] Size, anatomical location, and complications of the lesion are determinants of the treatment modality.[6]

Cystic hygroma at the cervical region may have accompanying tracheal deviation or upper airway obstruction depending on the degree of invasion to surrounding tissues that may contribute to the difficulty in airway management.[7]

Given its relative rarity, there is still no standard airway management technique for patients with cystic hygroma. Existing literature have described the use of intramuscular, intravenous, and inhalation induction as well as the use

of conventional laryngoscope, video laryngoscope, and fiberoptic scope.[7-12] This paper aims to describe an intravenous induction technique coupled with a novel way of using a nasopharyngeal airway to facilitate fiberoptic nasotracheal intubation in a child with cervical cystic hygroma.

CASE DESCRIPTION

Patient is a 4-year-old, 16 kg, 110 cm female diagnosed with cystic hygroma since birth. After being delivered vaginally, she was already noted to have soft movable mass on the right mandibular area with no accompanying respiratory distress. At two months old, ultrasound revealed 5.3 x 4.3 x 5.6 cm large, avascular, multiloculated, complex predominantly cystic right anterolateral neck mass which confirmed the diagnosis of cystic hygroma. She was lost to follow-up since then.

Patient was brought to ER due to sudden enlargement of neck mass associated with undocumented fever, cough, colds, dysphagia with sialorrhea, anorexia and dysphonia but with no dyspnea. Patient prefers to be on a side lying position with the mass on the dependent side while sleeping and was able to eat only soft food due to dysphagia. There was no note of stridor or use of accessory muscles. Physical examination showed a 14 x 10 x 9 cm soft, nontender mass on the right submandibular area extending to the submental area and right lateral neck. It was noted to be bulging from the right floor of the mouth which consequently pushed the tongue upward (Figures 1 and 2). Chest x-ray did not reveal any extension to the mediastinum (Figure 3). CT scan of the head and neck revealed multiple fluid-filled loculi in the anterior and right side of the neck with mild compression and deviation of the trachea to the left (Figure 4).

Aspiration of serosanguinous fluid was done which afforded slight relief of symptoms. A nasogastric tube was inserted for feeding. Patient was scheduled for prophylactic tracheostomy.

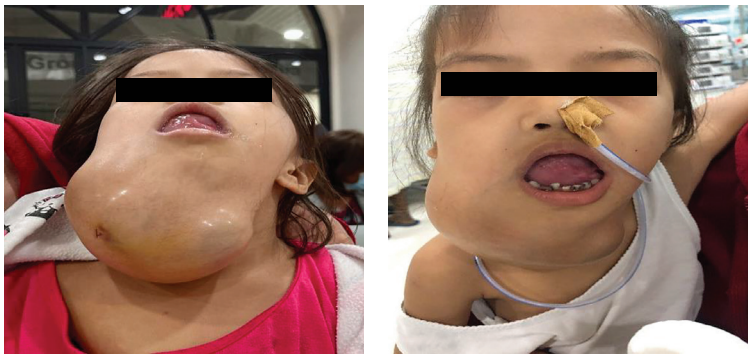

Figure 1. AP view of patient during neck extension and neutral position.

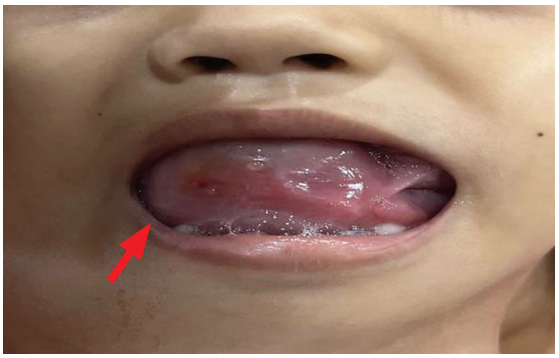

Figure 2. Patient with mouth wide open revealing extension of cystic hygroma *(red arrow)* in the oral cavity.

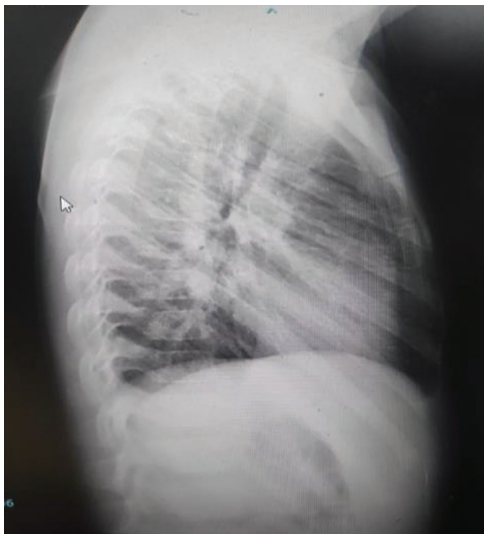

Figure 3. Lateral view of chest x-ray revealing normal chest findings.

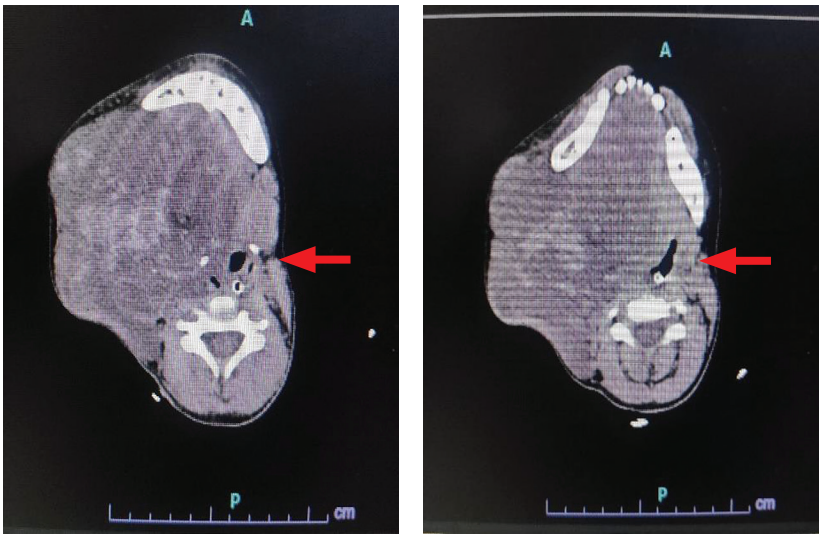

Figure 4. Neck CT scan axial view showing multiple cystic lesions with tracheal deviation *(red arrows)*.

Standard ASA monitors were attached upon arrival of the patient which showed BP 116/80 mmHg, HR 110 bpm, RR 26 cpm, SpO_2 98% at room air. Surgeons were on standby for possible emergency tracheostomy. Premedication consisting of atropine 0.2 mg IV and oxymetazoline hydrochloride 0.05% nasal spray per left nostril were administered. Intravenous sedation was achieved with midazolam 0.8 mg and ketamine 15 mg IV. Spontaneous ventilation was maintained with oxygen supplementation at 6 lpm via facemask. A well-lubricated pre-sliced nasopharyngeal airway French 26 (Rusch® adjustable flange Silkolatex ™ airway) from flange to bevel was inserted smoothly in the left nostril. Remifentanil infusion was started at 0.3 mcg/kg/min. A pediatric flexible fiberoptic scope with a preloaded cuffed north RAE size 4.0 ETT on the proximal end was inserted through the nasopharyngeal area inside the pre-sliced nasopharyngeal airway on the left nostril (Figure 5). Lidocaine 2% was given in a "spray as you go" technique through an epidural catheter placed along the fiberoptic scope until the tracheal rings and carina were seen through the scope. Upon visualization of the glottic opening, the nasopharyngeal airway was peeled off from the scope and the endotracheal tube was inserted (Figure 6). ETT was secured after confirmation of tube placement and depth of insertion with capnography and auscultation, respectively. Anesthesia was maintained with sevoflurane 3% and remifentanil infusion at 0.2 mcg/kg/min. The rest of the perioperative course was uneventful. Patient was referred for swallow evaluation, diet progression, and speech therapy. She was discharged on the 9th post-operative day and was scheduled for elective sclerotherapy c/o Interventional Radiology.

Figure 5. AP and lateral view simulation of how the flexible fiberoptic scope was inserted inside the pre-sliced nasopharyngeal airway.

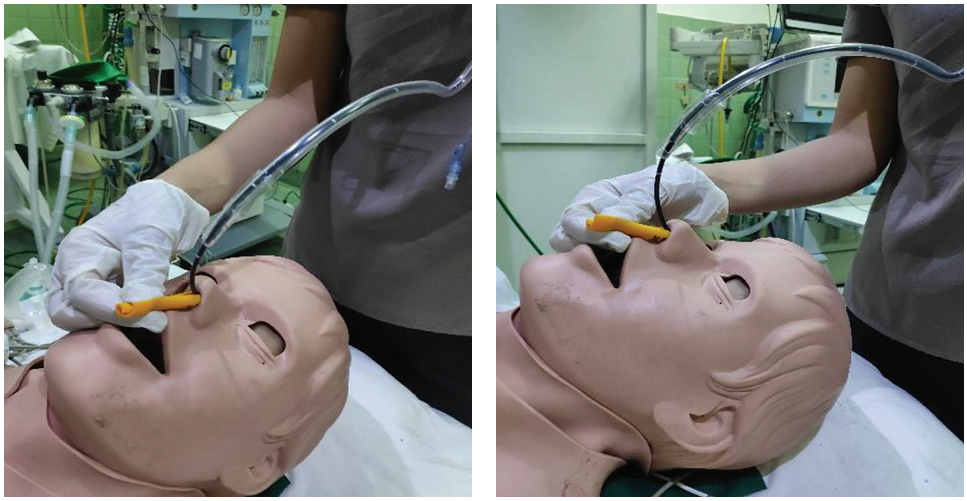

Figure 6. AP and lateral view simulation of how the pre-sliced nasopharyngeal airway was peeled off prior to inserting the ETT.

DISCUSSION

A benign lesion that is painless in nature, cystic hygroma can lead to sudden severe respiratory distress due to its propensity to proliferate and infiltrate surrounding tissues.[9,13-15] As it usually occurs among infants, an awake airway management approach that would have been customarily done in an adult with an anticipated difficult airway is not always a viable option and can only be performed after induction of anesthesia or deep sedation.[16,17]

The ideal induction technique is one that would allow tolerability of airway instrumentation while maintaining spontaneous respiration and upper airway tone. Inhalation induction has long been used among pediatric patients and has a good safety record. It is favored by others as it allows induction while preserving spontaneous respiration. It has been used successfully to induce patients with cystic hygroma.[7,9-11,14,15,18] However, maintaining a steady state level may be a limitation during airway instrumentation with this approach.

Apnea is a possible complication of intravenous induction agents.[15,19] This, however, is not a universal finding among all intravenous agents. Favorable results have been achieved with induction of cystic hygroma patients using ketamine, midazolam and ketamine combination as well as ketamine and dexmedetomidine combination.[8,12,13]

In the index patient, midazolam, ketamine, and remifentanil infusion were used to induce loss of consciousness and provide analgesia during intubation while maintaining spontaneous ventilation. Use of remifentanil infusion during the maintenance phase afforded sufficient sedation with rapid elimination of its effects upon infusion discontinuation.[20]

Direct and indirect laryngoscopy has been used successfully in these patients but it usually required more than one attempt.[7,10,13,18] Based on the PeDI registry, direct laryngoscopy is the most frequently (46%) attempted first tracheal intubation technique for difficult pediatric tracheal intubation encounters, however, its first attempt success rate is only 3%.[21]

There are documented cases with the successful use of video laryngoscope in these patients.[8,11] Although only 18% used it as the first tracheal intubation technique for difficult pediatric tracheal intubation encounters, it has a more favorable first attempt success rate of 55%.[21] It was not considered for this patient due to the significant extension in the oral cavity.

Another important concern is the possibility of trauma and bleeding due to possible frequent manipulation of the scope secondary to difficulty in visualizing the airway during the conduct of a fiberoptic-guided nasotracheal intubation. The nasal cavity is highly vascular. Hence, even minor external trauma to it can lead to epistaxis that may result to coughing, aspiration, and difficulty in visualizing the airway. Vasoconstrictor use, anti-sialagogue use, and lubrication are measures to prepare the nasal mucosa for the airway instrumentation.[22]

Nasopharyngeal airway is an airway adjunct used to maintain airway patency. It is not routinely used during the conduct of fiberoptic nasotracheal intubation. Holm-Knudsen described the use of a Portex Blueline ETT inserted in a nostril to serve as a nasopharyngeal airway both to maintain airway patency and maintain sevoflurane administration while a pediatric flexible fiberoptic scope is inserted in the opposite nostril among 19 children.[23] Low reported the use of a nasopharyngeal airway with tracheal tube connector to provide oxygen supplementation while fiberoptic nasotracheal intubation was performed via the opposite nostril.[24]

In the patient, a well-lubricated pre-sliced nasopharyngeal airway was inserted on the same nostril as the insertion site of flexible fiberoptic scope to serve as a scope passage guide and act as a protective device of the nasal cavity soft tissue and cystic mass during scoping while oxygen supplementation was maintained via a face mask. The insertion of the nasopharyngeal airway has also dilated the nostril further facilitating the insertion of the ETT.

Upon confirmation of intubation, the ETT is connected to the anesthesia machine and surgical anesthetic depth is safely maintained thereafter with balanced anesthetic technique using a volatile anesthetic and continuation of intravenous remifentanil infusion.

CONCLUSION

In a pre-school pediatric patient with anticipated difficult airway due to cystic hygroma, the use of a well-lubricated pre-sliced nasopharyngeal airway as a scope passage guide for flexible fiberoptic-guided nasotracheal intubation while receiving midazolam, ketamine and remifentanil infusion is a safe and effective airway management technique.

Informed Consent

Parent consent was obtained on the use of photographs and publication of the case.

Statement of Authorship

Both authors certified fulfillment of ICMJE authorship criteria.

Author Disclosure

Both authors declared no conflicts of interest.

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