

Anaesthetic management of a child posted for excision of lymphangioma of the tongue

Bhalla AP¹, MD, Mehta S¹, MD, DNB, Garg R¹, MD, DNB

ABSTRACT Lymphangioma is a congenital malformation of the lymphatic system, often involving areas of the head and neck. Patients may require surgical excision. Anaesthetic concerns include bleeding, difficulty visualising the airway, extrinsic and intrinsic pressure on the airway causing distortion, and enlarged upper respiratory structures, including the lips, tongue and epiglottis, which make airway management challenging. We report lymphangioma of the tongue in a six-year-old patient. There is limited information on the optimal anaesthetic management for this age group. The challenges with airway management, including bleeding, laryngospasm and difficult intubation, are outlined. Awareness of potential airway involvement and possible complications is necessary in order to provide safe anaesthesia to patients with lymphangioma.

Keywords: anaesthesia, airway, face mask, laryngoscope blade size, tongue lymphangioma
Singapore Med J 2012; 53(3): e45–e48

INTRODUCTION

Paediatric airways pose a challenge to anaesthesiologists if the airway pathology superimposes on an already potentially difficult airway. Lymphangioma is a congenital malformation of the lymphatic system, often involving areas of the head and neck.⁽¹⁾ The structures involved include an enlarged tongue and lips, swelling of the floor of the mouth and direct involvement of the upper respiratory tract. The definitive treatment for lymphangioma is surgery. Airway management in such patients is a nightmare for anaesthesiologists, especially in view of bleeding, difficulty in visualising the airway, extrinsic and intrinsic pressure on the airway causing distortion, and enlarged upper respiratory structures, including the lips, tongue and epiglottis. We report the peri-operative management of a case of large lymphangioma of the tongue that was scheduled for excision.

CASE REPORT

A six-year-old boy weighing 23 kg was scheduled for tongue lymphangioma excision. He presented at the paediatric surgery clinic with the chief complaint of a progressively increasing tongue mass since birth. The patient had difficulty eating, but there was no history of respiratory distress. He had a history of repeated chest infection, which was managed conservatively with antibiotics administration. Computed tomography revealed a lymphangioma involving the whole tongue, with massive enlargement of the anterior two-thirds of the tongue as well as adenoid enlargement.

On examination, the tongue mass was protruding out of the oral cavity. The mouth opening was around one-and-a-half fingers and mostly obliterated by the tongue mass (protruding out of the mouth), with Mallampati grade III (Figs. 1 & 2). Neck

movements (flexion and extension) were adequate and systemic examination was normal. Investigations revealed a haemoglobin level of 13.8 g/dL and a total leucocyte count of 8,900/mm³. Urine albumin and sugar were absent. Chest radiograph was normal. The child and his parents were counselled concerning the surgical procedure and fasting status. Premedication with syrup promethazine 12.5 mg in the morning was advised. Intramuscular injection of glycopyrrolate 0.2 mg was administered 45 minutes prior to the scheduled surgery.

On the day of surgery, the patient was moved to the operating room, and routine monitors, including electrocardiogram (lead II), pulse oximeter and noninvasive blood pressure monitor, were attached. After securing intravenous access, the patient was pre-oxygenated via the nose using an anatomical face mask (size 1) and adequate bag movement was observed. Anaesthesia was induced with intravenous ketamine 50 mg. Lungs were ventilated with 1% isoflurane in nitrous oxide and oxygen (50:50) via a face mask over the nose only, and the mouth was kept gently closed by an assistant. Oral laryngoscopy was attempted while the patient was spontaneously breathing using a Macintosh blade (size 3), which revealed Cormack-Lehane grade III; it improved to grade II after optimal external laryngeal manipulation. The trachea was intubated with an armoured cuffed endotracheal tube (internal diameter 5 mm).

After confirming the correct position of the tracheal tube with capnography and five-point auscultation, neuromuscular blockade was achieved with atracurium 15 mg intravenously. Anaesthesia was maintained with isoflurane in nitrous oxide and oxygen (MAC 1), along with boluses of atracurium. Analgesia was maintained with intravenous fentanyl (40 µg) administered after securing the airway. Rectal paracetamol suppository (500 mg)

¹Department of Anaesthesiology and Intensive Care, All India Institute of Medical Sciences, New Delhi, India

Correspondence: Dr Sunil Mehta, Senior Resident, Department of Anaesthesiology and Intensive Care, All India Institute of Medical Sciences, Ansari Nagar, New Delhi 110029, India. drrgarg@hotmail.com



Fig. 1 Photograph shows the child with a tongue lymphangioma protruding outside his mouth.

was also inserted at the start of surgery. Surgical excision of the mass proceeded uneventfully (Fig. 3). There was about 100 ml of blood loss, which was replaced with crystalloids. The surgery lasted for 45 minutes. Post surgery, residual neuromuscular blockade was reversed with neostigmine 1.5 mg and glycopyrrolate 0.2 mg intravenously. The trachea was extubated in the left lateral decubitus position after thorough oral suctioning, with the child fully awake, while ensuring adequate respiratory function and no obvious bleeding. He was then moved to the postoperative anaesthesia care unit for observation and nursed in a lateral position. Analgesia was maintained with rectal paracetamol suppository. The patient made an uneventful recovery.

DISCUSSION

The incidence of lymphangioma has been reported to be 1.2–2.8 per 1,000 newborns.⁽²⁾ Lymphangiomas are classified as microcystic (capillary lymphangiomas), macrocystic (cavernous lymphangiomas) and cystic hygromas according to the size of the lymphatic cavities incorporated.^(3,4) Lymphangiomas have a predilection for the head and neck region, which accounts for about 75% of all cases. About 50% of these lesions are noted at birth, and around 90% develop by two years of age.^(3,4) Airway obstruction occurs more frequently in newborns and young infants, although this tendency has not been statistically demonstrated.⁽⁵⁾ Lymphangiomas are known to be associated with Turner's syndrome, Noonan's syndrome, trisomies, cardiac anomalies, foetal hydrops, foetal alcohol syndrome and familial pterygium colli.

The various treatment modalities for lymphangioma are surgical excision, radiation therapy, cryotherapy, electrocautery,



Fig. 2 Pre-operative photograph shows the child with his mouth closed over the tongue lymphangioma.

sclerotherapy, steroid administration, embolisation and ligation, laser surgery with Nd:YAG and carbon dioxide, and radio-frequency tissue ablation technique; the treatment of choice is surgical excision.^(3,6,7) Our case highlights the potential anaesthetic implications of patients undergoing intubation with oropharyngeal lymphangioma. Most of the medical literature has focused on the surgical and airway management of this malformation at the time of birth. There is limited information on the optimal anaesthetic airway management for this age group.

The peri-operative concerns in our patient were regarding bag mask ventilation, tracheal intubation, oral vs. nasal intubation and history of chest infection. As in all potentially difficult airway cases, a thorough airway examination and interview with the family would aid in the complete assessment of the child. Parents may be able to answer questions relating to the patient's tendency to snore and ability to lie flat when sleeping. Snoring may indicate a predilection for airway obstruction, as the muscles of the tongue and pharynx relax during induction.⁽⁸⁾ Symptoms of dysphagia may indicate a decreased ability to protect the airway after extubation. A complete history of recent upper respiratory infections is also necessary, as respiratory infections may exacerbate the lymphangioma by causing oedema and enlargement.^(8,9) There are several characteristics of lymphangioma that need to be anticipated when providing anaesthesia.⁽⁸⁻¹¹⁾ The first concern is the extent to which the lymphangioma has affected the airway. Multiple surgeries and frequent infections in the airway can lead to scarring and fibrosis, which can further block lymphatic drainage.^(8,9) Although our patient had no previous surgery, his lymphangioma had affected the whole tongue, especially the anterior two-thirds of the tongue.

The difficult airway cart needs to be well-equipped before surgery. Lymphangioma and previous treatments can affect the patient's airway, necessitating the individualisation of airway management technique with a variety of oral airways and laryngoscope blades. We should ensure the availability of face masks, and nasal and oral airways of multiple sizes when anaesthetising such patients. A report of a nine-year-old child



Fig. 3 Postoperative photograph shows the child after excision of the tongue lymphangioma.

requiring a number nine adult oral airway due to the size of the base of his tongue has been documented.⁽⁵⁾ A larger-sized blade of laryngoscope may be required to negotiate an enlarged tongue and also to prevent injury during laryngoscope insertion. Sometimes, hardened airway structures due to disease pathology may be difficult to move from view during endotracheal intubation. We should ensure that a working suction machine is available during airway management in a child with dysphagia and be prepared for possible bleeding. Suction should be done gently, and if possible, under vision so as to avoid mucosal trauma. As tracheotomy is necessary in more than 50% of patients, a working suction machine is imperative in the operating theatre.⁽⁵⁾ Awake fibre-optic bronchoscope-assisted intubation remains the management of choice in such cases.

Sedative premedication is a major concern in patients with difficult airways. We administered promethazine to our patient, as he had no history of respiratory compromise and could lie comfortably in a supine position. We had an alternate plan for fibre-optic intubation in the event that bag and mask ventilation was not feasible and sedation for airway preparation was required. Therefore, anaesthesiologists and otorhinolaryngologists should be prepared to execute peri-operative airway management in such cases.⁽⁷⁾ The plan for induction of anaesthesia needs to be individualised, and intravenous vs. mask induction should be discussed prior to the procedure. Cooperation of the child and support of the parents are essential. Muscle relaxants should be used only after the airway has been secured due to a high probability of complete airway obstruction.

In our patient, macroglossia had made routine mask ventilation almost impossible. Intravenous induction was used, as we could

ventilate the lungs by keeping a small-sized mask over the nose and by closing the mouth over the protruded tongue. The oral seal could be adequately maintained in our patient, and lung ventilation was optimal with this technique. In an earlier reported case, a larger adult-sized face mask was used in a child with a tongue mass, allowing the enlarged tongue to be accommodated in the mask itself.⁽⁷⁾ In our case, we used a smaller-sized face mask for nasal ventilation. Before inducing anaesthesia, the child was asked to fully close the mouth with the tongue protruding outside and to breathe through the nose via a face mask. Adequate bag movements were confirmed. This technique has an added advantage; as the tongue remains outside and fixed at the mouth, it lowers the risk of the tongue falling back and causing respiratory obstruction.

A wide range of devices has been developed to aid in the management of difficult airways. They incorporate a variety of fibre-optic, video, optical and mechanical technologies to enable the operator to obtain a better view of the larynx as well as to facilitate the passage of an endotracheal tube into the trachea.^(11,12) Recently, in an attempt to propose a revised difficult airway algorithm, it was concluded that tracheal intubation can be achieved successfully in a large cohort of patients with a new management algorithm incorporating the use of gum elastic bougie, Airtraq and LMA CTrach™ devices.⁽¹³⁾ Awake intubation may not be easily performed in children since cooperation is imperative. Premedication and pre-oxygenation should be followed by inhalation of sevoflurane in a spontaneously breathing patient. Intubation should be performed under deep inhalational anaesthesia, but if this is difficult, the anaesthesiologist must have a secondary plan on how to proceed. In our patient, oral intubation was planned, as the patient had moderately enlarged adenoids, and thus, there was a high probability of injury to the adenoids and a possible risk of aspiration on nasal intubation. The size of the laryngoscope blade should be appropriately chosen according to the size of the lymphangioma so as to minimise injury during laryngoscopy.

In conclusion, anaesthesia for a child with lymphangioma involving the oral cavity and oropharynx requires preparation and vigilance. Knowledge of the disease and experience in previous surgeries and with affected airway structures would help the anaesthesiologist to ensure that all necessary equipment is available and to determine the safest plan for administering anaesthesia.

REFERENCES

1. Sichel JY, Udassin R, Gozal D, et al. OK-432 therapy for cervical lymphangioma. *Laryngoscope* 2004; 114:1805-9.
2. Jeeva Rathan J, Harsha Vardhan BG, Muthu MS, et al. Oral lymphangioma: A case report. *J Indian Soc Pedod Prev Dent* 2005; 23:185-9.
3. Filston HC. Hemangiomas, cystic hygromas and teratomas of the head and neck. *Semin Pediatr Surg* 1994; 3:147-59.
4. Harashima T, Hossain M, Walverde DA, Yamada Y, Matsumoto K. Treatment of lymphangioma with Nd: YAG laser irradiation: a case report. *J Clin Laser Med Surg* 2001; 19:189-91.

5. Hartl DM, Roger G, Denoyelle F, et al. Extensive lymphangioma presenting with upper airway obstruction. *Arch Otolaryngol Head Neck Surg* 2000; 126:1378-82.
6. Mikhail M, Kennedy R, Cramer B, Smith T. Sclerosing of recurrent lymphangioma using OK-432. *J Pediatr Surg* 2009; 30:1159-60.
7. Tewari A, Munjal M, Kamakshi, et al. Anaesthetic consideration in macroglossia due to lymphangioma of tongue: a case report. *Indian J Anaesth* 2009; 53:79-83.
8. Sharma S, Aminuldin AG, Azlan W. Cystic hygroma: anaesthetic considerations and review. *Singapore Med J* 1994; 35:529-31.
9. Cohen SR, Thompson JW. Lymphangiomas of the larynx in infants and children. A survey of pediatric lymphangioma. *Ann Otol Rhinol Laryngo Suppl* 1986; 127:1-20.
10. Goldberg MH, Nemarich AN, Danielson P. Lymphangioma of the tongue: medical and surgical therapy. *J Oral Surg* 1977; 35:841-4.
11. Thompson BM, Welna JO, Kasperbauer JL, Van Ess JM, Marienau ME. Childhood airway manifestations of lymphangioma: a case report. *AANA Journal* 2004; 72:280-3.
12. Kanai M, Horimoto Y, Yoshioka H, Fujino S, Takono T. [Perioperative management for partial resection of a lymphangioma of the tongue]. *Masui* 1996; 45:869-72. Japanese.
13. Amathieu R, Combes X, Abdi W, et al. An algorithm for difficult airway management, modified for modern optical devices (airtraq laryngoscope; LMA CTrach™): A 2-year prospective validation in patients for elective abdominal, gynaecologic and thyroid surgery. *Anesthesiology* 2011; 114:25-33.