Case Report

Novel Usage of Dexmedetomidine In A Paediatric Patient With Giant Tongue Haemangioma

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ABSTRACT
Giant haemangioma of the tongue is a disease which can obstruct the oropharyngeal airway and is presented with obstructive symptoms. Due to its vascularity, inserting laryngoscope for intubation can cause high risks, such as inducing bleeding. Hypoxia and excessive bleeding must be anticipated while securing the airway. We present a case of novel usage of dexmedetomidine as a conscious sedation agent for awake fibre optic intubation in a 9-year-old child with obstructive symptoms secondary to a huge tongue haemangioma, who was presented for interventional sclerotherapy of the lesion.

Keywords: Difficult airway, Tongue Haemangioma, Awake fibre optic intubation, Paediatric

INTRODUCTION
Haemangioma of the tongue is the most common abnormality of the vascular plexus formation. It originates since birth and can rapidly proliferate leading to various anatomical airway abnormalities (1). The patients are usually presented late with a complex airway because of the obstructed airway which is due to the tongue falling backwards or infiltration of the haemangioma tissue to the pharynx and larynx. Awake fibre optic intubation is a gold standard technique in anticipated problematic airways without upper airway obstruction. It is more difficult in children. Uncooperative children may add to the challenge leading to unsuccessful intubation. Trauma and bleeding might lead to asphyxiation. Traumatized tumour during airway manipulation may lead to uncontrolled bleeding. The chances of getting traumatized is more during the fibre optic intubation in the limited space of the airway associated with the huge tumour.

CASE REPORT
A 9-year-old, female child, a diagnosed case of tongue haemangioma was scheduled for sclerotherapy procedures by an interventional radiologist under general anaesthesia. She was diagnosed clinically since birth. However, she defaulted the treatment. She had a history of a rapidly enlarging tongue associated with dysphagia, snoring and breathlessness during lying supine, which made her constantly need to sleep in a lateral position. However, lack of sleep observation or overnight SpO2 monitoring was conducted with this patient. She was initially presented again at the age of 9 years old with minor bleeding from a small swelling of the tongue which had not caused any airway obstruction. She was planned for sclerosing therapy upon diagnosis. However, 6 months later she came with a huge tongue haemangioma which required major changes in the anaesthetic management of her condition. There were no episodes of cyanosis or excessive bleeding during sleep at this presentation. Examination revealed a very huge swelling of the protruded tongue haemangioma which completely occluded the airway (Figure 1). There was no radiological investigation performed to evaluate the extension of the haemangioma inferior and posteriorly prior to the procedure because evaluation of the haemangioma is an invasive procedure that requires sedation. Respiratory and cardiovascular systems examinations were unremarkable. Mouth opening was very limited, allowing only one-finger-breath. Mallampati score could not be assessed because of the compromised view. Preoperative laboratory tests including haematology parameters were normal. High risk consent was taken in view of the challenging airway, anticipating of respiratory problems during the post-operative period. She was planned for awake fibre optic intubation with dexmedetomidine infusion with the otorhinolaryngology team standing by for tracheostomy. An Intensive care unit (ICU) bed

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During this whole period, the SpO\textsubscript{2} remained above 95% with continuous oxygen supply via nasal prongs. Intravenous Dexamethasone 2 mg was given after intubation because of prolonged airway handling to prevent airway edema. Intravenous Dexamethasone 2 mg was given immediately followed by 10 mg of rocuronium. Anaesthesia was deepened with 1.5–2% sevoflurane in 100% oxygen. SpO\textsubscript{2} and End tidal CO\textsubscript{2} were constantly monitored. Fibre optic was rerouting via the endotracheal tube, which revealed ETT in situ 1 cm above the carina and anchored at 20 cm at nostril. Laryngeal and pharyngeal structures were not clearly visualized and intra-oral space was limited in huge tongue haemangioma during fibre optic scope. During this whole period, the SpO\textsubscript{2} remained above 95% with continuous oxygen supply via nasal prongs. Intravenous Dexamethasone 2 mg was given after intubation because of prolonged airway handling to prevent airway edema. Intravenous Dexamethasone 2 mg was given immediately followed by 10 mg of rocuronium. Anaesthesia was deepened with 1.5–2% sevoflurane in 100% oxygen. SpO\textsubscript{2} and End tidal CO\textsubscript{2} were constantly monitored. Fibre optic was rerouting via the endotracheal tube, which revealed ETT in situ 1 cm above the carina and anchored at 20 cm at nostril. Laryngeal and pharyngeal structures were not clearly visualized and intra-oral space was limited in huge tongue haemangioma during fibre optic scope. During this whole period, the SpO\textsubscript{2} remained above 95% with continuous oxygen supply via nasal prongs. Intravenous Dexamethasone 2 mg was given after intubation because of prolonged airway handling to prevent airway edema.

In the operation theatre, the equipment for awake fibre optic intubation for paediatrics were prepared and checked. Besides this, the difficult airway trolley was also prepared, which included different sizes of endotracheal tubes (ETT), laryngeal mask airways (LMA), and laryngoscope blades. Paediatric CMAC videolaryngoscope was also prepared. Intravenous (IV) access was obtained. Basic monitors including electrocardiogram, non-invasive blood pressure, and pulse oxymeter were attached to the patient. Nebulized lidocaine 2% 3 cc was given at the induction room. Then she was given glycopyrrolate 0.1 mg, and cocaine 4% 20 mg soaked in ribbon gauze to the more patent right nostril. IV dexametomidine loading 1 mcg/kg over 10 minutes followed by infusion 0.5 mcg/kg/hr as an adult dose was given. However, she was still uncooperative and needed the second loading dose of 1 mcg/kg over 10 mins and an increased infusion up to 2 mcg/kg/hr together with IV midazolam 0.3 mg and fentanyl 20 mcg titrated to maintain spontaneous respiration.

She was successfully intubated with fibre optic bronchoscope size 3.8 mm with ETT size 5.5 mm after two attempts. Intubation was confirmed with the presence of end tidal CO\textsubscript{2}. IV propofol 40 mg was given immediately followed by 10 mg of rocuronium. Anaesthesia was deepened with 1.5–2% sevoflurane in 100% oxygen. SpO\textsubscript{2} and End tidal CO\textsubscript{2} were constantly monitored. Fibre optic was rerouting via the endotracheal tube, which revealed ETT in situ 1 cm above the carina and anchored at 20 cm at nostril. Laryngeal and pharyngeal structures were not clearly visualized and intra-oral space was limited in huge tongue haemangioma during fibre optic scope. During this whole period, the SpO\textsubscript{2} remained above 95% with continuous oxygen supply via nasal prongs. Intravenous Dexamethasone 2 mg was given after intubation because of prolonged airway handling to prevent airway edema.

Anaesthesia was maintained with 2% sevoflurane with MAC 1 in oxygen-air mixture (50:50) with fentanyl 20 mcg/hr. IV propofol 3–4 mg/kg/hr were infused to maintain deep sedation during transportation to the radiological suite and was continued throughout the procedure to maintain deep anaesthesia. The patient was put on pressure-controlled ventilation. Peak airway pressures were around 17 cmH\textsubscript{2}O and the respiratory rate of 16/min were kept to achieve the end tidal CO\textsubscript{2} between 32 and 35 mmHg. For analgesia, fentanyl 60 mcg in titrating dose was given prior to the procedure. The sclerosing therapy procedure which was done by a radiologist, consisted of injecting mixtures of ethanol and lipid intravenously inside the haemangioma guided by a C-shape CT-scan. During the procedure, the diagnosis of haemangioma was confirmed by seeing the movement of the sclerosing agent inside the blood vessels using a C-shape CT-scan. It was successfully completed within 2 hours. However, she required a repeated procedure in view of the very huge haemangioma.

Post-operatively, the child was observed in the ICU in view of anticipating airway edema and the need for elective tracheostomy to avoid the risk of difficult intubation in the subsequent therapy. Paracetamol 200 mg IV was given every 6 hourly for post-operative analgesia. The child remained comfortable and pain free and did not have any respiratory difficulties. She then underwent elective tracheostomy prior to extubation for future re-sclerosing therapy. The tracheostomy was uneventful and she was extubated and discharged home with a scheduled appointment in 3 months.

**DISCUSSION**

Giant haemangioma of the tongue may present itself with a difficult airway especially in children. The chronic progressive course is caused by the proliferation and accumulation of abnormal blood vessels, causing swelling of tissue and compromising the airway. Disease presentation is usually at birth. Initial common symptoms include small red spots swelling over the tongue which then slowly increases in size. The patients are usually presented late with difficult airways because of tissue infiltration into the airways. Macroglossia is often the clinical findings.

Intravenous induction agents should be avoided to maintain spontaneous ventilation throughout the procedure (2). Local or regional anaesthetic techniques are unsuitable as the sole form of anaesthesia in these young children in view of the very painful procedure and the risk of aspiration if bleeding occurs throughout the procedure. Total intravenous anaesthesia techniques may be one of the options even though there is a risk of apnea with the induction agent. However, emergency intubation limits this option as the first choice of induction.

Inhalational induction has been used as a preferred technique because of the potential for airway obstruction (2). However, the inhalational agent was not suitable in this type of patient in view of
of difficulties with mask ventilation. Intravenous induction with ketamine or low-dose thiopentone maintains spontaneous ventilation, until definite airway control is achieved and has been used in uncooperative patients. However, this method will result in excessive airway secretion as the side effects of ketamine might obscure the view during laryngoscopy. Benzodiazepine, opioids, ketamine and dexmedetomidine are the main group of drugs that have been used to facilitate awake fibre optic intubation (3). We initially used dexmedetomidine bolus and infusion as a sole agent for intubation to maintain spontaneous respiration. Small doses of fentanyl and midazolam was used to supplement dexmedetomidine due to the uncooperativeness of the child.

Most patients require concomitant midazolam for procedural sedation in children despite dexmedetomidine infusion. Dexmedetomidine has sedative, analgesic and anxiolytic properties. It has been growing popular in paediatric anaesthesia and intensive care, even though it still hasn’t been fully studied in children. To obtain effective sedation, a larger boluses (2 to 3 mcg/kg/hr) and infusion rates (up to 2 mcg.kg.hr) of dexmedetomidine may be needed (4). However, at higher doses, it may cause more profound hypotension. Therefore, the usage of dexmedetomidine is required for titrating up to achieve the desired effect without causing complication to the patient.

We ensured adequate ventilation before giving muscle relaxant. Moreover, we used rocuronium, even after establishing ventilation, as patients can have dislodgement of the endotracheal tube which might need urgent reversal with sugammadex. There are cases of endotracheal dislodgment during manipulation of airway during surgery and transportation.

In anticipation of airway narrowing due to tissue infiltration, we first tried a 5.5 mm ID ETT, though 6.0 ID mm was the age appropriate sized ETT for this patient. Tracheal intubation was impossible with a 4.5 mm ID ETT in view of the size of the paediatric scope 3.8 mm and can only accommodate ETT 5 mm and above. The use of videolaryngoscope for managing difficult airways may provide better view of glottis but difficulty in negotiating it, still remains because of very limited space in the oral cavity in this case. In an emergency situation, emergency surgical tracheostomy is one of the choices applied to secure the airway. In the absence of otorhinolaryngologist, criocothyroidotomy must standby in case of failed intubation and requires emergency airway access. However, criocothyroidotomy is not recommended for children and adolescents in view of the risk of vocal cord damage (5). Therefore, it should be avoided in children and in the absence of surgeon. If required, it should be converted to surgical tracheostomy as soon as possible to reduce the risk of subglottic stenosis.

In conclusion, novel usage of dexmedetomidine as a sedation agent for awake fibre optic intubation was found successful in preserving spontaneous respiration throughout the procedure in a paediatric patient with a giant tongue haemangioma.

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