Dexmedetomidine-propofol for tubeless spontaneous respiration technique in an 8 week old infant with congenital subglottic cyst, laryngotracheomalacia and tracheal hemangioma

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Abstract
Laryngomalacia is a common cause of stridor in infants, however concomitant congenital subglottic cyst and tracheal hemangioma are both rare. We describe the management of an 8-week old infant presenting with three congenital airway lesions who underwent direct laryngoscopy and bronchoscopy under dexmedetomidine-propofol total intravenous anesthesia in a tubeless spontaneous respiration technique. 

Keywords: Infant, dexmedetomidine, propofol, laryngomalacia, laryngoscopy, bronchoscopy.

Introduction
Laryngomalacia is the most common cause of stridor with incidence reported between 1: 1500 and 1 : 2500. In a review of 130 patients by Adil et al, (1) concomitant congenital anomalies are common up to 48%. Subglottic and tracheal hemangioma have an incidence of ~1.5%. Therefore, the presence of laryngotracheomalacia with concurrent subglottic cyst and tracheal hemangioma in our patient is considered a rare clinical condition. Endotracheal intubation remains to be the standard approach and the most convenient method in securing the airway in children undergoing direct laryngoscopy and bronchoscopy (DLB). However ,the presence of an endotracheal tube and the small area of the glottic opening makes complete visualization of the airway difficult. There is great compromise between the surgeon and the anesthesiologist with regards to surgical manipulation and airway ventilation. Hence, a tubeless spontaneous respiration (TSR) technique with volatile inhalational agents supplemented with topical local anesthesia has been an alternative method. However a major drawback of this technique is the scavenging of volatile anesthetic gas leaks. Recent modification of TSR technique using propofol with inhalational gas has been studied (2). However, the issue of gas leaks and its effect to the surgeon and staff remains. The author describes combining an old technique with intravenous agents for...
laryngoscopy and bronchoscopy obviating the need for inhalational agents.

**Case report**

This is a case of an eight week old female weighing 4.8 kg, presenting with progressively increasing stridor. Patient was born full-term via cesarian section. She developed noisy breathing at 6 weeks, prominent on sleeping hours and during feeding. On admission, patient came in with inspiratory stridor, alar flaring, supraclavicular, substernal retractions, and oxygenation saturation at 93% with supplemental oxygen. The upper airway examination was normal. Arterial blood gas showed a compensated respiratory acidosis with hypoxemia. Initial impression was to consider tracheomalacia versus a subglottic stenosis. Chest x-ray revealed an atelectasis on the right lung with compensatory hyperaeration on the left lung. Videofluoroscopy done showed severe tracheal stenosis. CT scan with 3D reconstruction also revealed airway anomalies. On the 4th hospital day, patient was referred to anesthesia service with a plan to do a direct laryngoscopy with rigid bronchoscopy to ascertain the nature of the obstruction. Patient was risk stratified as ASA III. Initial anesthesia plan was general endotracheal anesthesia (GEA). However, due to uncertainty of the etiology, level and degree of stenosis, the plan was revised to total intravenous anesthesia (TIVA) without intubation and maintaining spontaneous respiration. The infant was then placed on standard fasting with no premedications given.

At the operating room (OR), atropine 100 mcg (20 mcg/kg) and midazolam 0.5 mg (0.1 mg/kg) IV was given. Patient was hooked to 100% oxygen via nasal cannula (OmniLine™) capable of end-tidal carbon dioxide (etCO₂) monitoring. Continuous infusion of Dexmedetomidine at 1 mcg/kg/hr was started before induction of anesthesia. After ten minutes, a bolus of Propofol at 10 mg (2 mg/kg) was given and a spray of lidocaine 1% applied to oropharyngeal area. Vital signs were noted to be stable with presence of spontaneous respiration at rate of 30-35 cycles per minute, oxygen saturation at 98-99% saturation and etCO₂ at 33-35 mmHg. Before the insertion of the rigid bronchoscope, another bolus of Propofol 10 mg (2 mg/kg) and Dexmedetomidine 3.5 mcg (0.7 mcg/kg) was given. A pediatric circuit with infant mask was made available for positive pressure manual bagging if she went to apnea or rapid desaturation. The ENT service was also prepared to do an emergency tracheostomy for definitive airway access. Examination revealed three areas of constrictions in the airway. The first is a smooth, round cystic mass at the left lateral subglottic wall, non-friable, obstructing approximately 90% of the area. The second constriction was a mass at the upper third of the trachea probably tracheomalacia versus granulation. The third constriction was a smooth, reddish mass with prominent vascularities on the lower 3rd of the trachea, just above the carina, at the 7 o’ clock position (Figures 1, 2, 3)

**Fig. 1.** Videofluoroscopy of patient at 8 weeks of age

**Fig. 2.** Patient at 8 weeks of age showing subglottic constriction; subglottic airway noted to have a smooth, round cystic mass at the left lateral subglottic wall, non-friable, obstructing approx. 90% of the area; note of a smooth, erythematous mass
Patient remained hemodynamically stable and no episodes of oxygen desaturation throughout the procedure which lasted 20 minutes. Postoperatively, she was continued on supplemental oxygen at 2 liters per minute via nasal cannula. Paracetamol oral drops 0.5 ml (10mg/ kg) for analgesia and dexamethasone 2.5 mg (0.5 mg/kg) IV. Patient was discharged, treated with beta blockers on an out patient basis and improved upon subsequent follow up consultations with main service.

**Discussion**

GEA with inhalational agents is the conventional anesthetic method for microlaryngoscopy and bronchoscopy. The main disadvantage of this technique is that it limits the visual field for examination. Second, sometimes it requires the use of relaxant which would restrain the surgeon’s assessment of airway patency and laryngeal dynamics. Third, anesthesia is not maintained in the desired level of sedation because of intermittent interruptions with ventilation. Lastly, the risk for acquired laryngeal stenosis and subglottic cyst is increased due to mechanical erosion and eventual scar formation. It was initially contemplated as the anesthetic plan in our patient, however, with the videofluoroscopic finding of severe stenosis there was uncertainty of whether patient could be intubated using an endotracheal tube with a wider internal diameter to fit the pediatric rigid bronchoscope (STORZ™) with an external diameter of 3.5 mm. Another concern was, if the endotracheal tube could be inserted safely without injuring the larynx or puncturing the prominent vascularities. Hence, a tubeless technique was employed.

Tubeless spontaneous respiration (TSR) was safely utilized. This technique depended on the patient’s normal respiration and offers the advantage of providing an unobstructed view of her larynx. Traditionally, TSR is done with anesthetic gas. However, gas leaks results in inadequate maintenance of minimum alveolar concentration and interrupted delivery of anesthesia plus OR room pollution. Hence, total intravenous agents were utilized. Some studies who have used TSR with inhalational-intravenous agents for microlaryngeal procedures in children reported difficulty in monitoring etCO2 reasoning only minimal increases of pCO2 level in spontaneously breathing patients(3). However, the use of a nasal cannula (OmniLine™) for end-tidal carbon dioxide (etCO2) monitoring, serves as a beneficial guide to detect the presence of adequate ventilation. This allowed better titration of anesthetic agents.

Dexmedetomidine infusion at 1mcg/kg/hr was started in the patient as a premedication. It provided anxiolysis, an arousable sedation without respiratory depression. It is able to maintain ventilation and airway patency even with increasing sedation level. It also obviates the need for opioids which can affect consistency of respiration. Propofol was one of the anesthetic used in the case due to its rapid onset and offset of action. It has been safely administered in pediatric population for sedation and maintenance of anesthesia. The use of propofol significantly reduced MAC requirements for halothane while not significantly affecting anesthetic and surgical outcome during TSR (2). No report of using propofol alone in TSR was found by the author. This might be due to the fact that children have higher requirements for propofol; and, if not titrated well to effect may cause apnea, rapid desaturation and subsequent, intubation. Intermittent bolus of propofol was preferred over infusion since it was shown that for short procedures it is both effective and efficient in maintaining the level of anesthesia. Although, propofol depresses airway reflexes and lowers oropharyngeal muscle tone, during emergence it normally results in a good airway (4). The addition of dexmedetomidine bolus on top of propofol bolus and dexmedetomidine infusion was done because it was found out to reduce tracheal reactivity (5). The need for general endotracheal anesthesia was totally obviated by employing total intravenous anesthesia using dexmedetomidine with propofol during TSR. However, since high doses of propofol causes apnea, the addition of dexmedetomidine was necessary for its ane-
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Sedation and reduction effect; also, for its sedative-analgesic
effect without respiratory depression since TSR rely
heavily on normal respiration for ventilation. No opioids
were given intra-operatively or postoperatively because
of its respiratory depressing effect.

Conclusion

The presented case demonstrates that dexmedetomidine-
propofol TSR seems to be an alternative to general
endotracheal intubation or inhalational anesthetic gas.
It provides the surgeon to have a better free access to the
larynx without the disadvantage operating room
pollution. Most importantly, it provides for a practical
and safe conduct of anesthesia without necessarily
depressing the respiration of the patient.

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