

CASE REPORT

Anesthesia for patients with subglottic cysts

Daniel N. Vo

Department of Anesthesiology, Perioperative, and Pain Medicine, Boston Children's Hospital, Harvard Medical School, 300 Longwood Ave, Bader 3, Boston, Massachusetts, 02115

Correspondence

Daniel N. Vo, Department of Anesthesiology, Perioperative, and Pain Medicine, Boston Children's Hospital, Harvard Medical School, 300 Longwood Ave, Bader 3, Boston, MA 02115. Tel: 857 218 5574; Fax: 617 630 0610; E-mail: daniel.vo@childrens.harvard.edu

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Key Clinical Message

Acquired subglottic cysts can cause rapid development of respiratory distress. Subglottic cysts are a disease of premature infants and other pathologies of prematurity should be anticipated. Perioperative success is dependent on communication between surgeon and anesthesiologist. Contingency plans for an emergency surgical airway should be in place in the event of total airway obstruction.

Keywords

Airway obstruction, pediatric ENT, pediatric otolaryngology, subglottic cysts.

Introduction

Subglottic cysts (SGCs) are a rare pediatric diagnosis with an estimated incidence of 1.9 per 100,000 births [1, 2]. They are of concern for the anesthesiologist because they can rapidly progress to severe respiratory distress and complete obstruction. Patients with subglottic cyst may require anesthesia for procedures such as flexible or rigid bronchoscopy and subglottic cyst excision. Here, we present a case of acquired subglottic cysts, discuss the natural history of SGCs, and makes recommendations for the perioperative anesthetic management of these patients.

Case

A 4-month-old ex-26 week male infant, weighing 5 kg, with a past medical history significant for bronchopulmonary dysplasia and chronic lung disease presented to the Emergency Department (ED) after rapidly developing stridor and respiratory distress. His parents denied any viral prodrome. In the ED, he was given albuterol and atrovent with minimal improvement. A fiberoptic laryngoscopy performed by an otolaryngologist showed mild laryngomalacia. He was discharged home on methylpred-

nisone with the diagnosis of viral upper respiratory infection and scheduled for follow-up with his pediatrician the next day. At home, the patient's condition worsened, prompting the parents to return to the ED. En route, they noted that he became quiet and cyanotic at which point they pulled over and called 911.

In the ED, the patient was started on continuous albuterol nebulizers and given a loading dose of terbutaline. A venous blood gas at that time revealed a pH of 7.27 and PCO₂ of 55. He was admitted to the PICU and his respiratory support was escalated to CPAP of 5 and eventually to BiPAP of 17/7.

In the PICU, the pulmonologist noted biphasic stridor worsening with agitation on exam. The absence of wheezing on exam made him question the diagnosis of reactive airway disease, and he recommended discontinuing the albuterol for concern of causing airway collapse in patients with bronchomalacia. He also recommended an emergent direct laryngoscopy and rigid bronchoscopy under general anesthesia be performed.

On the day of the procedure, the patient was maintained on BiPAP *en route* to the OR. After receiving 1 mg/kg of ketamine via an *in situ* IV, he was transitioned to a face mask with CPAP and gentle bag-mask

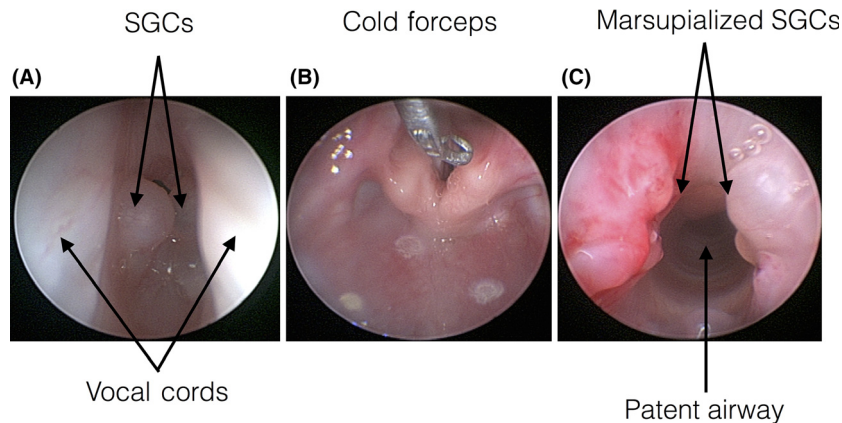


Figure 1. (A) Preoperative subglottic cysts. (B) Marsupialization with cold steel instruments. (C) Post-operative appearance of the airway.

assistance. The patient was allowed to spontaneously breathe Sevoflurane. The patient's vocal cords were topicalized with lidocaine via an atomizer. On rigid bronchoscopy, the patient was found to have enlarged subglottic cysts occluding >90% of his trachea (Fig. 1A). The cysts were marsupialized with cold steel instruments, and the patient was intermittently masked with sevoflurane. After cyst resection, the patient had dramatic improvement in air entry and was returned to the ICU spontaneously ventilating via a natural airway and was discharged after 3 days.

Discussion

Subglottic cysts are classically a disease of preterm infants. The typical history is a low-birth weight infant born prematurely with respiratory distress requiring intubation and management in the NICU [3]. In one series, over 90% of patients had a gestational age of 25–31 weeks [4]. Duration of intubation was usually several days (mean: 10–20 days) [3, 4]. Time to development of cysts ranged widely from days to years with an average of 5 months [3, 4]. The diagnosis of SGCs appears to be increasing, which could be due to better survival of preterm infants, improved anesthetic techniques, and improved scopes [5]. The mechanism of SGC formation is unknown, but postulated to be iatrogenic phenomena caused by the repeated piston trauma of the endotracheal tube and the ventilator [6]. The subglottic region is the narrowest portion of the pediatric airway. Subglottic ducts are present in greater number in infants as compared to adults. Injury and scarring of the subglottic mucosa is thought to lead to blockage of the ducts and cyst formation [7].

Preoperative assessment of these patients should determine the urgency and severity of symptoms. A birth history should seek out other pathologies of prematurity. One should seek out the diagnosis of bronchopulmonary

dysplasia, chronic lung disease, or reactive airway disease. Prior anesthetic records can indicate the number and duration of previous intubations as well as any history of difficult intubations. Home medications should be reviewed for bronchodilators, recent steroid courses, and diuretic use. Baseline oxygen saturations as well as home oxygen therapy or ventilator support should also be clarified. Patients with severe symptoms or comorbidities (e.g., lung disease or congenital heart disease) should have a preinduction IV placed.

Preanesthetic preparation for airway procedures requires close communication between the anesthesiologist and otolaryngologist. Intervening on the airway while maintaining adequate oxygenation and ventilation requires the cooperation of two distinct airway specialists with differing priorities and skill sets. A mutual respect and understanding of each specialty's goals allows for optimizing surgical conditions while maintaining patient safety. The discussion should include the surgeon's preference for examining the airway under spontaneous ventilation or with muscle relaxation. Contingency plans including a plan for emergency tracheostomy in the event of difficult ventilation or intubation should be delineated.

There are multiple approaches to safely anesthetizing the patient with subglottic cysts. Premedication can help calm patients and avoid worsening stridor. Patients requiring ventilatory support should have it maintained until induction of anesthesia in the OR. Even with a preoperative IV, a combination induction (inhaled with IV agent) is useful for providing positive airway pressure, controlling the speed of induction, maintaining spontaneous ventilation, and monitoring of airway patency. Once the patient is induced, topicalization of the vocal cords under direct laryngoscopy with a Laryngeal Tracheal Anesthesia (LTA) device accomplishes two goals: blunting airway reflexes and assessing depth of anesthesia before proceeding with rigid bronchoscopy.

Maintenance of anesthesia can be accomplished with volatile agent or TIVA. TIVA with spontaneous ventilation using propofol and remifentanyl has been described by Ferrari et al. [8]. Advantages are that it provides a tubeless, spontaneously ventilating patient reducing interruptions to the procedure. It also avoids contaminating the room with potent agents. The drawbacks are the added time required for titrating infusions to the narrow window of spontaneous ventilation and blunted airway reflexes. If TIVA is used, a dedicated, visible peripheral IV is desirable. In the absence of end-tidal CO₂ monitoring, a precordial stethoscope is extremely useful for determining the quality of ventilation.

Maintenance with volatile agent requires intermittent masking or intubation. While masking, the stomach should be inspected for inadvertent insufflation. If intermittently intubating, an undersized, uncuffed ETT can reduce trauma and postoperative laryngeal edema. Volatile anesthetic cannot be delivered while the patient is extubated, requiring intermittent delivery of high concentrations at high flows. Typically, the hemodynamic consequences of this are well tolerated in pediatric patients, but may not be suitable in patients with impaired preload, SVR, or cardiac reserve. Apneic oxygenation via insufflation of supplemental O₂ into the oropharynx increases the P_AO₂ and delays desaturation. Pediatric patients have roughly twice the oxygen consumption of adults and continue to accumulate CO₂ during apnea. This approach may not be suitable for patients with pulmonary or intracranial hypertension.

The gold standard therapy is endoscopic marsupialization with either cold steel instruments or CO₂ laser [3]. Once the cysts have been marsupialized, the improvement is often dramatic. After the procedure, the patient can be intubated for emergence or recovered via face mask. If there has been extensive manipulation of the airway, the patient should remain intubated and monitored in the ICU for the development airway edema. Most children require a repeat endoscopic procedure for reassessment. Recurrence has been estimated anywhere from 20% to 60% [5, 6] though this seems to be less with the use of cold instruments [9]. As the incidence of this pathology seems

to be increasing, the anesthesiologist may encounter these patients more frequently in the OR. Their anesthetics can be performed in a variety of ways as long as attention is paid to adequate ventilation, communication between services, and the presence of contingency plans.

Conflict of Interest

None declared.

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