Airway malacia in premature infant twins with bronchopulmonary dysplasia: Two case reports

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Abstract
Premature infants who require surgery for retinopathy of prematurity often exhibit bronchopulmonary dysplasia. Reactive airway is a clinical manifestation of bronchopulmonary dysplasia. We describe premature infant twins who had difficulty with positive pressure ventilation during anesthesia. Both cases occurred during induction of anesthesia for binocular anterior chamber puncture and vitreous cavity injection. The most likely cause in each case was airway malacia. We recommend that endotracheal intubation is performed in infant patients with low body weight; the possibility of airway malacia occurrence should be considered, especially for infants with comorbid bronchopulmonary dysplasia.

KEYWORDS
airway malacia, bronchopulmonary dysplasia, retinopathy of prematurity

1 | INTRODUCTION

Retinopathy of prematurity (ROP) is a potentially blinding condition that typically affects preterm neonates with low gestational age and low birth weight.\(^1\) ROP is a common disease in premature infants and requires surgical treatment as soon as possible. Patients usually undergo sedation or general anesthesia, which may not be safe in systemically unstable infants.\(^2\) Anesthesia induction is particularly challenging because these patients may have many comorbid diseases. Here, we describe difficulty in positive pressure ventilation during anesthesia induction in premature infant twins diagnosed with bronchopulmonary dysplasia (BPD). The most likely cause was airway malacia.

2 | CASE PRESENTATION

Two premature twin boys were born at 28 weeks' gestation and weighed 1100 g and 1000 g, respectively. Their Apgar scores were both 10 at 1 min and 5 min after birth. However, poractant alfa was intratracheally administered for shortness of breath shortly after birth. Noninvasive mechanical ventilation was subsequently required during the first days of life because of respiratory distress syndrome; severe BPD was ultimately diagnosed because of inhaling 40% O\(_2\). Both infants were diagnosed with anemia, hypoalbuminemia, congenital patent foramen ovale, congenital hypothyroidism neonatal sepsis, and ROP in our hospital when they were transferred at 34 weeks' gestation. They were scheduled to undergo binocular anterior
chamber puncture and vitreous cavity injection for ROP under general anesthesia.

2.1 | Case 1

The older twin was scheduled to undergo the operation at the age of 1 month and 14 days (corrected gestational age, 35 weeks; weight, 1900 g). This patient was diagnosed with congenital cytomegalovirus infection, which was not present in the younger twin. He had discontinued ventilator use for approximately 2 weeks before the operation. Preoperative evaluation revealed a fair overall condition (heart rate, 150 beats/min; saturation of pulse oxygen (SpO₂), 99% under supplemental O₂). However, SpO₂ temporarily decreased to 70%–80% during bottle-feeding, according to neonatal intensive care unit (NICU) records. Electrocardiogram and chest radiography findings were generally normal.

In the operating room, the patient’s SpO₂ was 100% under O₂ (3 L/min via face mask), systolic blood pressure was 70 mm Hg, and heart rate was 160 beats/min. General anesthesia was induced by inhalation of 8 vol% sevoflurane. The induction process demonstrated difficulty during positive pressure ventilation with a face mask when the patient’s breathing was weakened, and airway resistance was increased. SpO₂ decreased to 90% under FiO₂ 1.0, and a laryngeal mask (1.0") was immediately placed. Wheezing was evident on lung auscultation in both lung fields. SpO₂ increased gradually to 100%; the wheezing was incompletely resolved after treatment with adrenaline and salbutamol. Under pressure-controlled ventilation, the patient’s tidal volume was approximately 20 ml at a pressure of 20 mm Hg; his respiratory rate was 30 times/min, inhalation concentration was 4–5 vol% sevoflurane, and end-tidal carbon dioxide was 30 mm Hg.

The operation duration was 30 min; anesthesia time was 1 h 32 min. After discontinuation of sevoflurane, spontaneous respiration was recovered and the laryngeal mask was removed. However, difficulty during positive pressure ventilation with a face mask was observed twice and SpO₂ decreased to 50% at the lowest point, despite treatment with 10 µg adrenaline, but SpO₂ maintained 100% soon after a 3.0" endotracheal tube applied. At last, the patient was observed for half an hour and transferred to the NICU with endotracheal intubation, considering the potential for airway malacia. After 4 h, extubation was performed and supplemental O₂ (4 L/min) was continuously administered via nasal cannula. The patient was discharged 10 days later.

2.2 | Case 2

Ten days later, the younger twin underwent a similar operation (age, 1 month and 24 days; weight, 2,300 g). Similarly, SpO₂ temporarily decreased to 70%–80% during bottle-feeding. Notably, this patient had discontinued ventilator use for only 1 day before the operation. He also exhibited severe anemia (75 g/L hemoglobin). Moreover, he was examined for presumed congenital patent foramen ovale (2 mm) and bidirectional atrial shunt. Considering the intraoperative complications experienced by his twin, endotracheal intubation was selected by the anesthesiologists. As expected, difficulty with positive pressure ventilation occurred during the induction process. Thus, a 3.0" endotracheal tube was immediately applied. The patient completed a 30-min operation without further complications. Extubation was performed 1 h later in the NICU; he was discharged 3 days later.

Both patients underwent second operations at corrected gestational ages of 43 and 48 weeks, respectively. The remaining surgical and anesthesia processes did not involve further complications.

3 | DISCUSSION

Sudden difficulty in positive pressure ventilation after general anesthesia induction can originate from various factors including inadequate anesthesia depth, tracheomalacia, airway malacia, and tongue suffix. In both cases, desaturation developed during manual mask ventilation under 8 vol% sevoflurane before tracheal intubation. Because these patients were twins, we presumed that they were physiologically similar in terms of growth and development. Furthermore, both patients underwent general anesthesia induction by the same anesthesiologists 2 months later, using identical inhalation anesthesia methods. No complications occurred during the second operations. Therefore, we considered the potential for a non–anesthesia-related cause. An alternative explanation for the desaturation event could be possible airway malacia in infants with dysontogenesis, that would explain the temporary reduction of SpO₂ during bottle-feeding; it would also explain the successful outcomes of the second operation and anesthesia in both patients, 2 months later.

Airway malacia can occur in the larynx (laryngomalacia), trachea (tracheomalacia), or bronchi (bronchomalacia). As a group, these are the most common congenital abnormalities of the pediatric airway and are characterized by increased airway compliance, resulting in excessive dynamic collapse during the respiratory...
cycle. Laryngomalacia is the most common cause of stridor in newborns. Affected patients may present with noisy breathing, a classic high-pitched inspiratory stridor that worsens with feeding, and tracheomalacia is a rare congenital defect, characterized by the immaturity of the cartilaginous rings (usually at the distal third of the trachea), which leads to a weakness of the entire tracheal structure. It is more frequent in premature babies and can be associated with laryngomalacia. While a diagnosis can be suspected based on clinical history and physical examination, definitive evaluation is based on nasopharyngolaryngoscopy and/or bronchoscopy. In these two cases, we did not perform preoperative bronchoscopy and laryngoscopy, but the pulse oxygen will decrease when the infants drink milk and the respiratory symptoms during the operation, relationships between airway malacia and comorbid diseases of premature infants were strongly suspected. There is no definite guideline for diagnosis of airway malacia in premature infants during the first 4 months of life. However, many former infants with BPD who underwent ophthalmic surgeries in our institute did not experience difficulty during positive pressure ventilation.

BPD, a chronic lung disease of infancy, occurs in 40% of very low-birth-weight infants; the incidence of BPD increases as birth weight decreases below 1500 g. The assessment of BPD is conducted at 28 days postnatally or 36 weeks postmenstrual age on the basis of supplemental O₂ requirement. The disorder is classified as mild, moderate, or severe, according to gestational age and degree of supplemental O₂ requirement. Clinical features of BPD include tachypnea, intercostal retraction, increased airway resistance, wheezing or coarse crackle, and reactive airway.

Our findings in these cases suggest that airway malacia can cause or aggravate positive pressure ventilation difficulty in premature infants with BPD, although a causal relationship cannot be conclusively established. We recommend that clinicians consider the possibility of airway malacia occurrence in infants with low body weight, especially in the presence of comorbid BPD. If possible, the anesthesiologist should recommend a bronchoscopy and confirm the diagnosis based on the infant’s symptoms.

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CONFLICT OF INTEREST
The authors report no conflict of interest.

AUTHOR CONTRIBUTIONS
YWW and LDX were the patient’s anesthetists, reviewed the literature, and contributed to manuscript revision; ZHP and ZJX reviewed the literature and contributed to manuscript drafting; all authors issued final approval for the version to be submitted.

ETHICAL APPROVAL
The patient’s parent provided signed informed consent to the publication of the case. All the procedures performed in this study were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

CONSENT
Written informed consent was obtained from the patient to publish this report in accordance with the journal’s patient consent policy in online proofing system.

DATA AVAILABILITY STATEMENT
Data sharing is not applicable as no new data were generated.

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