Perioperative Use of High-frequency Oscillation Immediately after Birth in Two Neonates with Congenital Cystic Adenomatoid Malformation

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Congenital cystic adenomatoid malformation of the lung (CCAM) is a rare pulmonary malformation, often associated with pulmonary hypoplasia due to compression by cysts.1,2 The neonate with CCAM may develop acute and progressive respiratory distress because of emphysematous enlargement accompanied by ball-valve air entrapment by cysts. Conventional mechanical ventilation (CMV) may cause further expansion of the involved lobe. High-frequency oscillation (HFO), which is associated with reduced peak airway pressure, may have the advantage of being able to ventilate the lungs of the neonate with CCAM. We report two fetuses prenatally diagnosed as having CCAM who were successfully managed with HFO.

CASE REPORTS

Case 1. A 27-yr-old woman, gravida 2, para 1, was evaluated by prenatal ultrasound examination at 27 weeks’ gestation. Ultrasound of the fetus revealed a multicystic mass in the left fetal hemithorax with mediastinal shift to the right. The thoracic lesion was considered to represent CCAM or congenital diaphragmatic hernia. In view of the immediate need for neonatal resuscitation and surgery because of pulmonary hypoplasia, it was decided to perform cesarean delivery. At 38 weeks’ gestation, the patient underwent elective cesarean section under spinal anesthesia. To control maternal gastric acidity, the H2-receptor antagonist famotidine, 20 mg, was administered orally at bedtime and intravenously 1 h before operation. The patient was given diazepam 0.3 mg/kg intravenously to sedate the infant in order to prevent crying after birth, since crying might lead to further deterioration because of air entrapment.3 Within 10 min after diazepam administration, the patient delivered a 3,081-g boy, who cried weakly.

The trachea of the infant was intubated immediately after birth without mask ventilation, and his lungs were ventilated with HFO (Hummingbird BMO-20N) at a frequency of 15 Hz, a mean airway pressure of 15 cmH2O, stroke volume of 15 ml, and 100% oxygen. His Apgar scores were 5 and 8 at 1 and 5 min, respectively, and the diagnosis of CCAM in the left chest was confirmed on chest x-ray. Initial preductal arterial blood gases at a fractional inspired oxygen concentration (FiO2) of 1.0 were pH 7.32, carbon dioxide tension (Paco2) 49 mmHg, and oxygen tension (PaO2) 71 mmHg. Five minutes later, postductal arterial blood gases were pH 7.23, Paco2 48 mmHg, and PaO2 100 mmHg. Anesthesia was induced with pancuronium 0.5 mg and fentanyl 30 µg and maintained with increments of fentanyl to a total dose of 65 µg·kg⁻¹ with supplemental intravenous pancuronium. A left upper lobectomy was performed. During surgery, there was no evidence of the cyst enlargement. Postductal arterial blood gas data obtained after the left upper lobectomy showed pH 7.42, Paco2 28 mmHg, PaO2 346 mmHg on HFO at a frequency 15 Hz, a mean airway pressure of 11 cmH2O, and FiO2 1.0.

The neonate was transported to the intensive care unit (ICU). Eight hours later FiO2 was decreased to 0.9, and 2 h later to 0.8. Postductal arterial blood gases revealed pH 7.50, Paco2 48 mmHg, and PaO2 60 mmHg, suggesting persistent fetal circulation (PFC). Increasing FiO2 to 1.0 and mean airway pressure to 15 cmH2O resulted in an improvement in postductal arterial blood gases, to pH 7.41, Paco2 53 mmHg, and PaO2 275 mmHg. Mean airway pressure was decreased to 13 cmH2O 3 h later, and to 11 cmH2O another 2 h later. On the 3rd ICU day we gradually reduced FiO2 and removed the tracheal tube after a 6-day course of HFO. The infant was tachypnic for about 2 h after tracheal extubation but subsequently has been doing well.

Case 2. A 30-yr-old woman, gravida 3, para 2, was evaluated by prenatal ultrasound at 25 weeks’ gestation. Ultrasound demonstrated a multiple cystic area in the fetal right thorax with a lefward mediastinal shift and polyhydramnios, consistent with a macrocystic CCAM. At 30 weeks’ gestation, ultrasound revealed fetal hydrops, ascites, pleural effusion, and edema of the scalp. In an attempt to decompress the normal lung tissue and permit adequate lung growth, fetal transthoracic aspiration of the cysts under ultrasound guidance was performed at 30 and 31 weeks’ gestation under continuous epidural anesthesia. Fetal heart rate and uterine activity were monitored continuously, and the tocolytic agent ritodrine was infused continuously to prevent preterm delivery. At 32 weeks’ gestation, ultrasound showed a decrease in amniotic fluid. Evidence of ascites, pleural effusions, and edema of scalp had disappeared. The patient was given betamethasone to induce fetal lung maturation.

At 34 weeks’ gestation the patient underwent elective cesarean section under epidural anesthesia. Fentanyl 20 mg was administered orally at bedtime and intravenously 1 h before operation. She was given diazepam 0.3 mg/kg intravenously to prevent the infant from crying. Five minutes later, a 2,780-g girl was delivered.

The trachea of the infant was intubated immediately after birth. The 1-and 5-min Apgar scores were 8 and 9, respectively. The infant’s lungs were ventilated with HFO at a frequency of 15 Hz, a mean airway pressure of 15 cmH2O, stroke volume of 15 ml, and 100% oxygen; preductal arterial blood gases showed pH 7.20, Paco2 59 mmHg, and PaO2 325 mmHg. After radiologic diagnosis, anesthesia was induced with 15 µg fentanyl and 0.5 mg pancuronium and maintained with increments of intravenous fentanyl to a total dose of 74 µg·kg⁻¹ with supplemental intravenous pancuronium. The infant underwent resection of the CCAM that had occupied the right lower lobe. During surgery there was no evidence of enlargement of the cysts. At the end of the operation, postductal arterial blood gas analysis

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revealed pH 7.50, PaCO\(_2\) 24 mmHg, and PaO\(_2\) 278 mmHg on HFO at a frequency of 15 Hz, a mean airway pressure of 15 cmH\(_2\)O, and F\(_{\text{I}}O\(_2\) 0.9.

After transport to the ICU, preductal arterial blood gases showed pH 7.45, PaCO\(_2\) 22 mmHg, and PaO\(_2\) 382 mmHg, but postductal arterial blood gases showed pH 7.42, PaCO\(_2\) 23 mmHg, and PaO\(_2\) 94 mmHg, suggesting PFC. F\(_{\text{I}}O\(_2\) was increased to 1.0, leading to an improvement in postductal arterial blood gases. Transient PFC episodes were reversed by manual ventilation during periods of acute deterioration. On the 3rd ICU day, HFO was switched to CMV. On the 4th ICU day, F\(_{\text{I}}O\(_2\) was decreased gradually. The infant required mechanical ventilatory support for 8 days postoperatively, and the trachea was extubated on the 9th ICU day. Because of atelectasis of the right upper and middle lobe, the infant’s trachea was reintubated 2 days later. Her lungs were ventilated with CMV and her trachea was extubated successfully on the 24th postoperative day.

**DISCUSSION**

CCAM is a rare pulmonary malformation that usually involves a part of a lung lobe. Large malformation may lead to mediastinal shift and compression of the heart. Compression of the opposite lung may result in pulmonary hypoplasia. CCAM may result in stillbirth or neonatal death or in acute progressive respiratory distress.\(^1^,2\)

A crying, struggling neonate may trap substantial amounts of air during inspiratory efforts. If air entrapment continues, the cystic malformation then undergoes progressive enlargement as a result of the ball-valve effect due to abnormal or deficient bronchial cartilage. In our two cases, CCAM was diagnosed prenatally, and cesarean delivery, neonatal resuscitation, and surgery were planned. Diazepam crosses the placenta rapidly, and furthermore, the equilibrium fetal–maternal blood concentration ratio is close to unity 3–12 min after injection in the mother.\(^4\) Therefore, we selected diazepam for fetal anesthesia, and the 5–10 min for the interval from diazepam injection to the mother to delivery was considered appropriate. The neonates cried weakly after delivery, and immediately their tracheas could be intubated without prior mask ventilation. The major risk of diazepam injection is pulmonary aspiration of the mother. To reduce the risk, both mothers fasted for a minimum of 8 h before surgery and were premedicated with the \(H_3\)-receptor antagonist famotidine orally on the evening prior to surgery and intravenously 1 h prior surgery. Especially in the case of a full stomach without any preanesthetic preparation, rapid-sequence induction of general anesthesia may be better than heavy sedation when fetal anesthesia is required.

The lungs of the neonates immediately were ventilated with HFO, and there was no further clinical deterioration during surgery. In the management of the neonate with CCAM, HFO may have several advantages over CMV, as follows.

First, HFO is associated with lower peak airway pressures for any given level of mean airway pressure; this lower peak pressure reduces the possibility of air entrapment and barotrauma, since it is not only the absolute value of peak pressure during HFO but also the smaller difference between peak and mean pressures that is of significance.\(^5^,6\) Positive-pressure ventilation and positive end-expiratory pressure should be minimized to prevent further expansion of the involved lobe.\(^7\) HFO is superior to high-frequency “jet” ventilation because of the minimal risk of air entrapment with HFO.\(^8\) In our cases there was no evidence of enlargement of cysts.

Second, HFO can achieve hyperventilation with low peak airway pressure, which is helpful for decreasing the pulmonary vascular resistance. HFO has been used successfully in neonates with congenital diaphragmatic hernia for treatment of PFC.\(^9^,10\) In our cases, transient PFC developed postoperatively. The primary strategy for treatment of PFC is hyperventilation at the lowest possible mean airway pressure to minimize pulmonary vascular resistance. However, the attempt to reduce mean airway pressure must be balanced against the need to keep alveoli sufficiently expanded to avoid atelectasis and the resulting hypoxemia due to shunting. We started HFO at a mean airway pressure of 15 cmH\(_2\)O and decreased it to 11 cmH\(_2\)O since the child had adequate blood gases in case 1.

Third, HFO can be used safely during surgical procedures to maintain a relatively quiet surgical field during thoracotomy.

We preferred elective cesarean delivery in the operating room to enable us to provide fetal anesthesia and to prepare for resuscitation, surgery, and the application of HFO. For these reasons, we believe that cesarean delivery is preferable to vaginal delivery when CCAM is diagnosed prenatally. In this setting, a team approach is necessary to include maternal transport and fetal therapy in utero. Finally, in addition to planned delivery and immediate resuscitation and surgery, fetal anesthesia and HFO may be useful in the treatment of the neonate with CCAM.

**REFERENCES**

Postoperative Myocardial Ischemia Possibly Masked by Epidural Fentanyl Analgesia

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Epidural and intrathecal opioid administration has proven to be an effective analgesic therapy for a variety of painful states. These include cancer, the postoperative period, the first stage of labor, posttrauma, and myocardial infarction.† This report describes a patient with a history of coronary artery disease who had asymptomatic myocardial ischemia while being treated with a continuous epidural infusion of fentanyl for postcholecystectomy pain. It is postulated that the epidural fentanyl infusion may have masked the pain of myocardial ischemia.

CASE REPORT

An 85-yr-old, 66-kg man was admitted to the hospital with a 1-week history of right upper quadrant abdominal pain. A diagnosis of acute cholecystitis was made. Despite treatment with antibiotics he did not improve and was scheduled for cholecystectomy.

The patient had an 8-yr history of coronary artery disease with exertional angina and on four prior occasions had suffered subendocardial infarctions, the last of which occurred 23 months prior to admission. On each of these occasions he had substernal chest pain with radiation to the left arm. During each subendocardial infarction, his electrocardiogram (ECG) showed temporary ST-segment depression and T-wave inversion in leads V2–V4 without progression to Q waves (fig. 1). His creatine phosphokinase (CPK) MB isoenzyme fraction was mildly increased with each episode. Prior to the incident reported here, he had had angina once or twice per month and had been walking 2–3 miles each day, using sublingual nitrates prior to exertion. His past medical history was otherwise unremarkable. He had no history of diabetes mellitus. His admission ECG revealed a first degree atrioventricular (AV) block and slightly reduced voltage.


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