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CASE REPORT

ANAESTHETIC MANAGEMENT OF A NEONATE WITH CONGENITAL CYST ADENOMATOID MALFORMATION

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ABSTRACT

Background: Congenital cyst adenomatoid malformation (CCAM) is a rare congenital malformation of the lung, more common in males. Removal of the cyst (thoracotomy) during early age is the standard method of the treatment.

Case characteristic: We report the anaesthetic management of a 5 days old male neonate, weighing 1.9 kg undergoing thoracotomy under general anaesthesia.

Observation/Intervention: Intraoperative anaesthetic course was uneventful with all parameters within normal range except transient desaturation (70 %) during lung compression and it was managed with temporary removal of self retaining retractor. **Outcome:** Postoperative course was uneventful.Patient Congenital cyst adenomatoid malformation (CCAM) is a rare congenital malformation of the lung,more common was discharged home on the 10thpostoperative day.

INTRODUCTION

CCAM is a congenital anomaly of the lung resulting from abnormal fetal lung development. It is a rare congenital malformation of the lung representing 25% of congenital lung malformations and 95% of congenital lung lesions.[1, 2]This lesion occurs more often in males (1.8:1), and is primarily unilateral, but may occur bilaterally. [3]

Case History

The baby was born prematurely at 32 weeks, weighing 1.9 kg, and with an antenatal diagnosis of congenital cystadenomatoid malformation (CCAM). Following normal vaginal delivery, the child developed respiratory distress requiring ventilation and was transferred to NICU for further management. Both X-ray (Fig. 1) and computerized tomography (CT) scan of chest (Fig.2) showed large lesion32 x 33 x 56 mm (AP x TR x CC) in size in left lower lobe.

The baby was admitted to the NICU, and blood investigations were within normal limits. The baby was shifted to Operation Theater. On examination, he was breathing spontaneously, active and SpO_2 was 98% with oxygen supplements at 4 l/min nasal cannula. Auscultation of the chest did not elicit any abnormality. He had heart rate 140/min, blood pressure 82/40 mmHg, RR 50/min. The baby was wrapped in warm cotton wool gamgees and placed on the heating mattress. Standard monitors were applied. Ryle's tube was aspirated with a syringe. Peripheral IV access was secured. Pre oxygenation was done with 100 % oxygen for 3minutesvia Jackson Rees circuit and then gradually sevoflurane was started. The baby was premedication with inj. glycopyrolate 0.004 mg/kg and inj. ondensetrone 0.15 mg/kg intravenously. The percentage of sevoflurane was slowly increased up to 3 to 3.5 %, keeping close watch on the heart rate and respiration. Occasional gentle assistance was given. Once the baby reached the deeper plane of anaesthesia, sevoflurane was stopped and 2-3 breaths of 100 % oxygen were allowed. After introducing laryngoscope, a portex uncuffed endotracheal tube (No-3) was inserted. The baby did well with induction. After confirming air entry and proper fixation of ETT, the baby was kept in right lateral position for left thoracotomy. Anaesthesia was maintained with100 % oxygen and 1 to 1.5 % sevoflurane concentrations, on spontaneous breathing. Till the chest was opened, the baby was breathing spontaneously, about 3 to 4 liters oxygen with 1 to 1.5 % sevoflurane. Occasional gentle manual assistance was given. After the chest and the plura were opened, the baby was paralyzed with inj. atracurium 0.5 mg/kg. The maintenance dose of atracurium was a quarter of the initial dose. Fluid was given at the rate of 10 ml/kg/hr in the form of isolyte M. Blood loss was also monitored. Intraoperative anaesthetic course was uneventful with all parameters within normal range except transient desaturation (70 %) during lung compression and it

was managed with temporary removal of self retaining retractor.

Before the extubation, the baby was allowed to breathe air for 5-10 minutes maintaining its expiratory blast, heart rate and reflex activities.

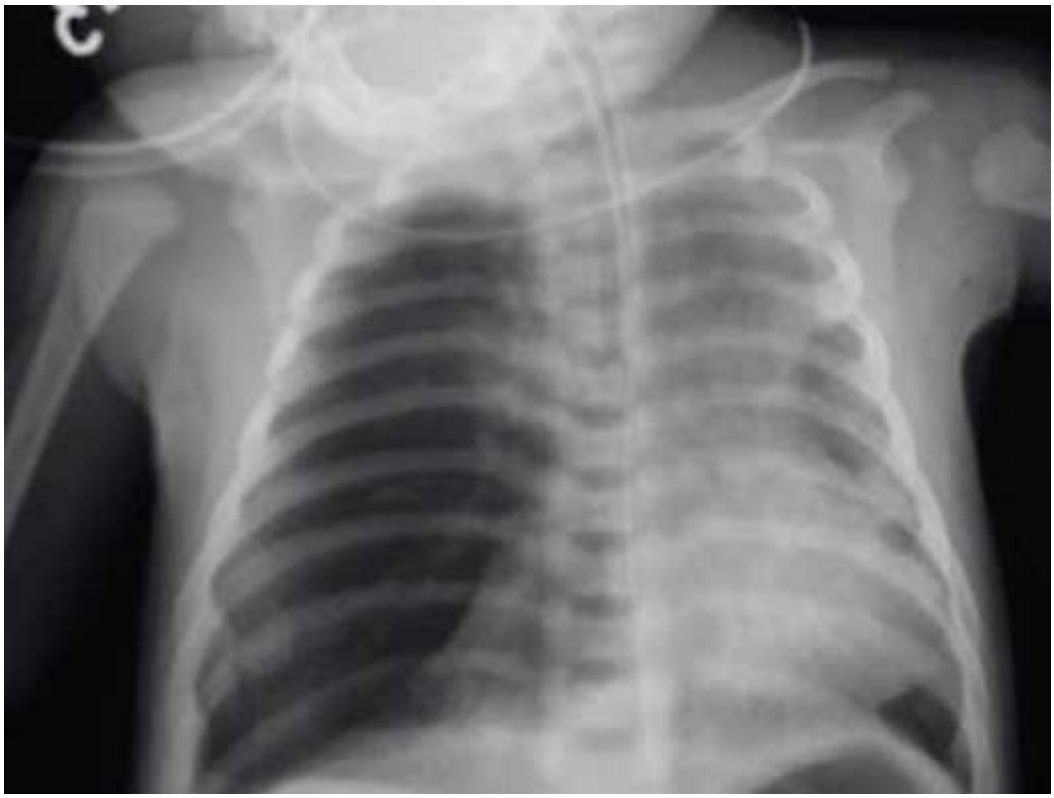

Figure 1 X-ray chest shows patchy segmental consolidation is seen in left lower lobe.

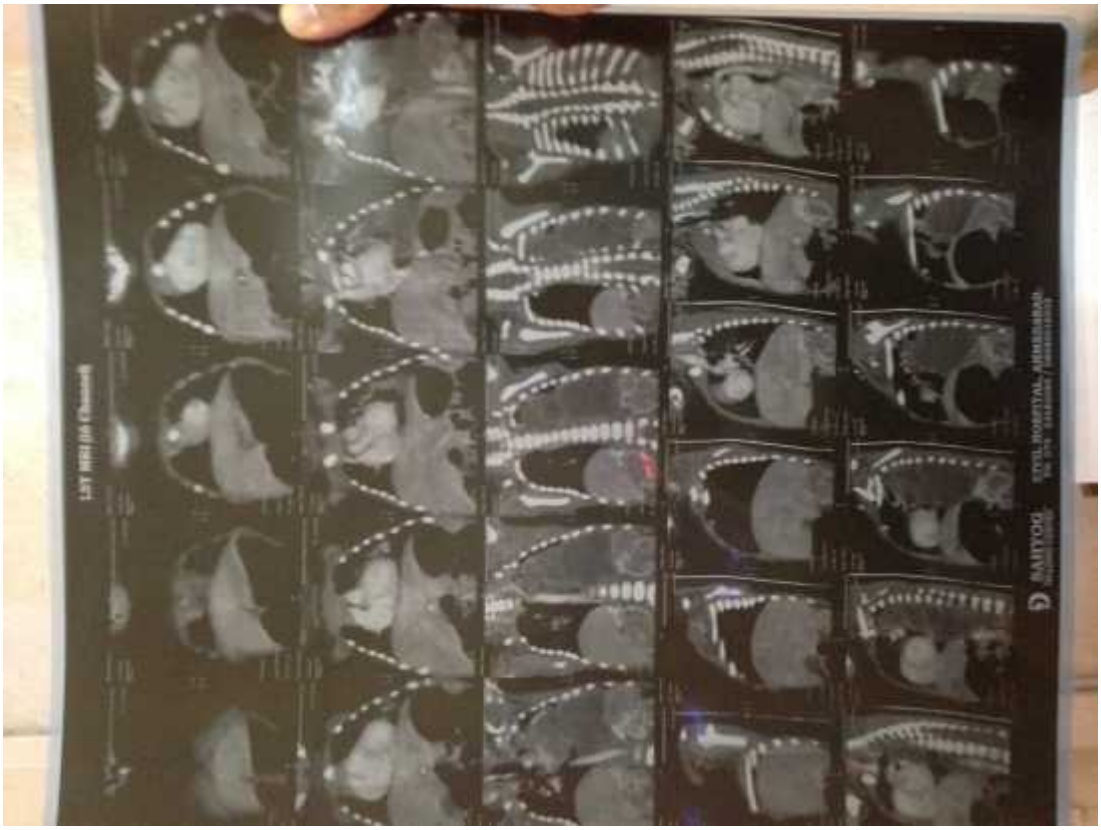

Figure 2 Computerized tomography (CT) scan of chest (Fig.2) showed large lesion 32 x 33 x 56 mm (AP x TR x CC) in size in left lower lobe.

The muscle relaxant effect was reversed with inj. glycopyrolate 0.008 mg/kg and inj. neostigmine 0.05 mg/kg intravenously.

The ETT was removed and given oxygen by a face mask, observed for 10-15 minutes and then shifted to NICU. Two liters of oxygen was given through oxygen tent and

temperature was maintained with a spotlight. Follow-up chest X-ray showed adequate expansion of the remaining lobe.

DISCUSSION

CCAM is a congenital anomaly of the lung resulting from abnormal fetal lung development. There is increased cell proliferation but decreased apoptosis, resulting in adenomatoid proliferation and cyst formation. The lesion is connected to the airway, but a normal intrapulmonary bronchial system is missing.

CCAM is the second most common congenital lung lesionl in children. The reported incidence of CCAM is 1: 11,000 live births. Higher incidence of CCAM is in mid trimester. CCAM occurs during branching and proliferation of bronchial structures. This lesion occurs more often in males (1.8:1), and is primarily unilateral, but may occur bilaterally. [3]

Associated anomalies are rare. Since the technological advancement of ultrasound examination, CCAM has been increasingly diagnosed on routine prenatal examinations. Some CCAM lesions present only at birth with respiratory distress symptoms but are confirmed by an abnormal chest radiograph or a more definitive computed tomography scan.

Monitoring of the vital parameters, during neonatal surgery is a must. During thoracotomy, the baby is at great risk.

On induction of anaesthesia if positive pressure ventilation is applied before opening the chest, it may cause rapid inflation of emphysematous lobe or cyst with sudden mediastinal shift and cardiac arrest.[7]Therefore, induction of anaesthesia should provide adequate spontaneous ventilation with minimal airway pressure. Occasional gentle assistance is necessary. Once the chest is opened and the affected lobe is delivered, the patient can be paralyzed and the lungs ventilated by controlled ventilation. [8]

Hyperinflation of the emphysematous lobe or cyst can be prevented by avoiding the use of nitrous oxide (N_2O) before the delivery of the affected lobe as nitrous oxide (N_2O) diffuses faster in closed cavity and expands the cavity, leading to further compression of normal lung and more mediastinal shift. [9]

In our case we used sevoflurane with spontaneous breathing till the chest and plura were opened, but we did not use nitrous oxide during the procedure.

Takrouri MSM *et al*. found that inhalation of sevoflurane (6%) for long period (3 minutes), patients did not relax enough for intubation and supplementation with ketamine and rocuronium are required to secure the airway. They concluded that intravenous induction with anaesthetic and muscle relaxant would control the airway. Also, gentle ventilation would provide oxygenation and elimination of carbon dioxide. [4]
Nishimoto C *et al*. found that one lung ventilation was necessary due to endotracheal tube became nearlyoccluded with copious purulent material during operative period. Theyconcluded anaesthesiologists should choose proper airway management method depending on the nature of cystic fluid. [5]

Tominaga H *et al*. was reported, intravenous induction with propofol and rocuronium without one lung ventilation for thoracotomy in neonate with CCAM.[6]

One lung ventilation, remifentanyl infusion, and ultrasound guided thoracic caudal epidural is an effective technique in neonates undergoing major thoracic surgery. [10]

We used sevoflurane with spontaneous breathing without nitrous oxide because of its potential for cyst expansion.

CONCLUSION

Congenital cyst adenomatoid malformation is rare congenital anomaly of the lung in neonate. The anaesthesiologist should choose proper airway management along with skillful perioperative anaesthetic method during thoracotomy in neonate.

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