

Case Report

Anaesthetic management of a neonate with congenital cyst adenoid malformation

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We report the anaesthetic management of a male neonate with congenital cyst adenoid malformation (CCAM) of the lung who underwent thoracotomy for resection of CCAM 24 h after birth and again at 24 days. The initial operation involved a 10-day admission to a paediatric intensive care unit (PICU) requiring ventilation, and was complicated by a pneumothorax. This report concentrates on the anaesthetic management for the second thoracotomy. The combination of intra-operative remifentanyl infusion and the use of ultrasound to confirm correct placement of epidural catheter allowed on-table tracheal extubation and a shorter stay in PICU. The use of one-lung ventilation (OLV) allowed for better surgical access and enabled complete resection of the lesion.

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The patient was born prematurely at 34 weeks, weighing 2.9 kg, and with an antenatal diagnosis of congenital cyst adenoid malformation (CCAM). Following normal vaginal delivery, the child developed respiratory distress requiring ventilation and was transferred to PICU for further management. Both an X-ray (Fig. 1) and computerized tomography (CT) scan of chest could not delineate the extent of the CCAM mass or identify normal lung tissue.

At 24 h the first operation was performed. Anaesthetic management included tracheal intubation and ventilation, arterial line, femoral central venous line, muscle relaxation, maintenance with isoflurane in an air/oxygen mix and morphine for analgesia. Intra-operative blood loss was more than 40% of total blood volume and required transfusion. Intra-operative findings were a single lung with a huge central CCAM mass occupying 70–80% of lung volume, with poor differentiation of the lungs into lobes. Because of blood loss and difficulty in distinguishing normal and abnormal lung only a small amount of the CCAM mass was removed. A chest drain was inserted at the end of the operation.

The child was ventilated in PICU with a morphine infusion for sedation/analgesia and required a blood transfusion overnight, but was otherwise stable. Complications included air leaks and pneumothorax drained by a chest drain. The trachea was extubated on the 10th postoperative day and the patient discharged to the high dependency unit

(HDU). The trachea had to be reintubated 4 h later and he was transferred back to PICU. On the 12th postoperative day the trachea was extubated and the child transferred to HDU on nasal CPAP. This was maintained until 24 days of age, when increasing requirement for respiratory support indicated the need for further surgery.

For the second thoracotomy standard monitoring was instituted and anaesthesia was induced with sevoflurane 8% in oxygen 100%. Peripheral i.v. access was secured and 1 mg kg⁻¹ rocuronium administered. At laryngoscopy, a size 3 Fr Fogarty catheter (guide wire in place) modified by bending the tip by 10°, was positioned in the trachea. The trachea was intubated with a 3.0 Vygon tracheal tube (with distal tip capnography). With fiberoptic guidance the tip of the Fogarty catheter was passed into the right main bronchus. The balloon of the Fogarty catheter was inflated under direct vision. The Fogarty catheter was then secured to the tracheal tube. The left radial artery was cannulated.

The child was placed in left lateral position for caudal catheter insertion. We used a Sonosite 180 Plus ultrasound machine with L38 10 MHz probe (in sterile sheath) to image the sacral hiatus, sacrococcygeal membrane, dura mater, and conus medularis. The estimated distance from the sacral hiatus to T5/6 level was 15 cm. Under aseptic conditions the sacral hiatus was palpated and an 18 G short Jelco (Medex Medical Ltd) i.v. cannula was inserted into the epidural

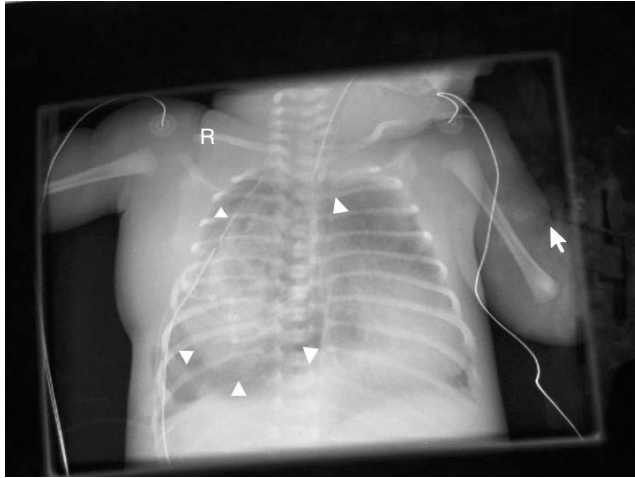


Fig 1 Chest X-ray of the neonate on first postoperative day after first thoracotomy. CCAM is seen as haziness in right hemithorax (within the markers) with mediastinal shift.

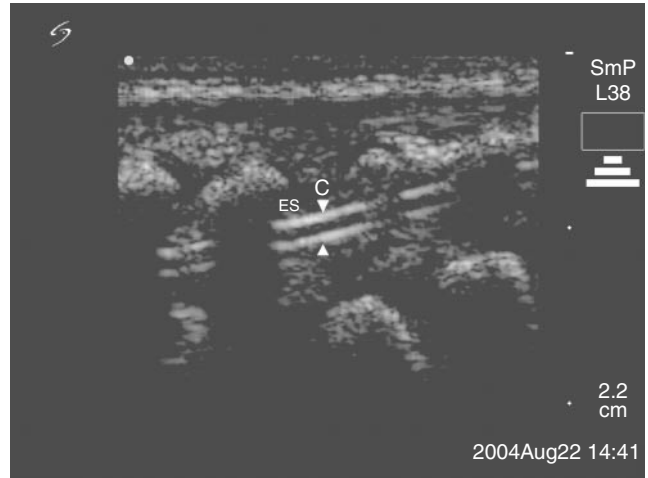


Fig 2 Ultrasound of caudal space with cannula (C) *in situ* seen as two parallel echo dense lines (within markers) in the epidural space (ES).

space. The position of the cannula within the space was confirmed using ultrasound (Fig. 2) to record the anterior displacement of the duramater following injection of 2 ml of saline test bolus. We used the catheter from a standard 18 G Portex epidural kit preflushed with saline. The epidural catheter was threaded up to 20 cm. Using an ultrasound probe placed in the midline at the level of T6 in the sagittal plane, the position of the epidural catheter was inferred from observing dural displacement on injection of saline through the catheter. Dural displacement was identified more cephalad than T5/6 and the echo-dense epidural catheter could be seen at this level. This was confirmed by gently moving the catheter back and forth and it was slowly withdrawn until the tip came into view, and was positioned opposite T5/6 (Fig. 3). Levobupivacaine 0.25% (1.5 ml) was injected and both caudad and cephalad spread observed on ultrasound.

Anaesthesia was maintained with sevoflurane (end tidal 0.7) in an air/oxygen mix, remifentanyl infusion (0.1–0.15 $\mu\text{g kg}^{-1} \text{min}^{-1}$), and epidural infusion (levobupivacaine 0.125% at 0.9 ml h⁻¹). The child received 20 ml kg⁻¹ of saline 0.45% + dextrose 2.5%, 20 ml kg⁻¹ of human albumin 4.5% solution and 20 ml kg⁻¹ of blood. The child remained haemodynamically stable. The bronchial blocker was inflated during the start of the operation and deflated for the final 40 min of the operation to allow detection of air leaks. The E'_{CO_2} rose from 5.3 to 7.7 kPa when the bronchial blocker was inflated for about 30 min and returned to 5.3 kPa when the blocker was deflated. Remifentanyl and sevoflurane were stopped at the start of skin closure. The operation time was 2 h 30 min. Neuromuscular block was reversed and spontaneous breathing with good tidal volumes returned 5 min later. At 25 min the child was awake and the trachea was extubated.

At this second operation the CCAM mass was resected completely and identified as arising from the right upper lobe. The left lung appeared small and hypoplastic. A

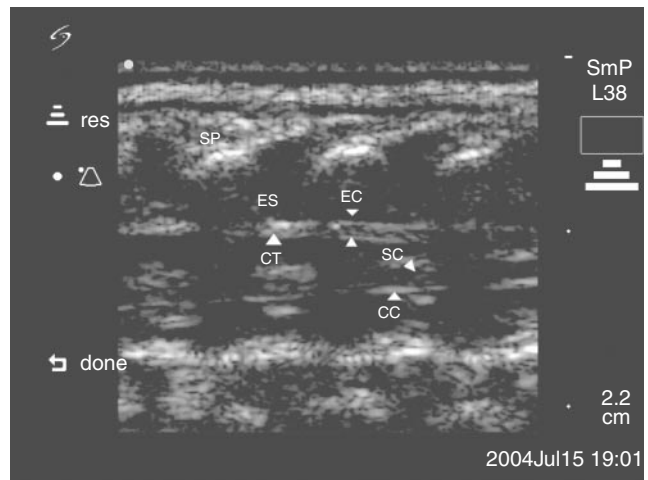


Fig 3 Ultrasound of thoracic epidural space showing catheter and tip of catheter in space. Structures: SP, spinous process; ES, epidural space; EC, epidural catheter; CT, catheter tip; SC, spinal cord; CC, central canal.

chest drain was inserted. The child was monitored in PICU overnight and required nasal CPAP for the first 2 h. Thereafter 0.5 litre min⁻¹ of oxygen by nasal cannula maintained satisfactory oxygenation. The following day the child maintained good respiratory function on air and was transferred to HDU, where the chest drain was removed 24 h later. The epidural was continued for 48 h at the rate of 0.3 ml kg⁻¹ h⁻¹. After epidural removal the child returned to the ward and was discharged home after 6 days.

Discussion

CCAM is the second most common congenital lung lesion¹ in children. It is more common in males and is the result of an embryologic insult before the 50th day of gestation causing maldevelopment of the terminal bronchiolar structures. This usually occurs in a single lobe causing ipsilateral lung compression, pulmonary hypoplasia, and occasional mediastinal shift. It can be diagnosed *in utero* by ultrasound

between 18 and 36 weeks and may disappear before birth. These children commonly die in the neonatal period and, hence, an urgent operation is needed to prevent further lung hypoplasia.

The management of a 24-day-old baby is different from that for a 1-day-old as the lesion is more mature and the physiology and pathology is well defined. One-lung ventilation (OLV) improves surgical access and may reduce blood loss. CCAM may contain fluid varying from clear to purulent in nature. At the first thoracotomy, resection of the lesion had been impossible with positive pressure ventilation of both lungs. OLV allowed better surgical access, minimized trauma to the limited residual normal lung tissue,² and protected normal lung from contralateral contamination. We used a 3 F Fogarty catheter as a bronchial blocker,³ which was placed parallel to the ETT. Alternatives include a balloon tipped angiography catheter or a 5 F paediatric bronchial blocker (Cook). Both can be passed into the bronchus using a guide wire and have the advantage of having an end hole allowing suction and collapse of the lung. The Cook blocker has the added advantage of a high volume low-pressure balloon. In small infants, the advantage may be offset by the greater diameter of the device. We could have also used a normal tracheal tube to intubate the normal lung alone but the disadvantages of doing this are difficulty in suctioning of secretions of the non-ventilated lung and obstruction of the left upper lobe bronchus.

Our plan was to extubate this child early to avoid positive pressure ventilation and the possible complications of barotrauma encountered after the first operation. Provision of good analgesia was therefore a priority. Studies in children suggest an epidural technique is better than morphine.^{4,5} We decided to insert an epidural catheter⁵⁻⁹ for analgesia because our patient had a stormy postoperative period after first thoracotomy. Bosenberg's work in 240 neonates undergoing major neonatal surgery showed significant benefit from epidural analgesia.⁵ We inserted a thoracic caudal catheter under ultrasound guidance as we had successfully used this technique in two previous cases. Incorrect positioning of a caudal cannula is common; the ultrasound saline test bolus confirms epidural position and rules out intravascular, intraosseous, or intrathecal placement. Ultrasound imaging allowed us to accurately position the catheter tip at the level of surgery and monitor the distribution of levobupivacaine. Other methods of confirming placement of epidural catheter have included ECG guidance (Tsui method⁷), electrical nerve stimulation and X-ray.⁹

Levobupivacaine is used for regional techniques in our institution, at a rate of 0.1–0.3 ml kg⁻¹ h⁻¹ for 48 h.

We used remifentanyl for its MAC sparing effect. Nitrous oxide was not used because of its potential for cyst expansion. Previous reports on the management of CCAM have not used remifentanyl, and analgesia has been provided by epidural⁷ or morphine infusion. All patients required prolonged postoperative ventilation. Patients who have had postoperative ventilatory problems have been tried on high-frequency oscillation¹⁰ and extracorporeal membrane oxygenation has been used, as pulmonary hypertension can be a major cause of morbidity and mortality.¹¹

In summary, OLV, remifentanyl infusion, and ultrasound-guided thoracic caudal epidural is an effective technique in neonates undergoing major thoracic surgery.

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