

Perioperative Anesthetic Care of Children with Congenital Cystic Adenomatous Malformation: A Report of 3 Cases

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ABSTRACT

Congenital cystic adenomatoid malformation (CCAM) is a rare developmental entity. Symptomatic patients must undergo thoracic surgery for lobectomy. Perioperative anesthetic management is challenging because it involves thoracotomy in a young patient that may lead to hemodynamic compromise and inadequate ventilation. We present 3 cases of pediatric patients (6 months - 2 years old) with CCAM who underwent lobectomy.

Keywords: congenital cystic adenomatoid malformation, pediatric, thoracic surgery, tracheal extubation, lobectomy

INTRODUCTION

Congenital cystic adenomatoid malformation (CCAM) is a rare developmental malformation affecting the lower respiratory tract. The reported incidence is 1 per 8,300 to 35,000 live births.^{1,2} It is characterized by multi-cystic areas of over proliferation and dilatation arising from tracheal, bronchial, bronchiolar, or alveolar tissue. It does not participate in gas exchange. Large cystic lesions compress the adjacent normal lung tissues. CCAMs occur sporadically, and there is no genetic predisposition. They are usually unilobed and unilateral, with arterial supply and venous drainage from the pulmonary circulation.

The postnatal management of symptomatic CCAM is well established and varies on whether the patient presents with respiratory distress or is asymptomatic. The decision between surgical management and observation is controversial for patients who remain asymptomatic.^{3,4} Adequate treatment of symptomatic patients requires a lobectomy or pneumonectomy in cases of extensive multi-lobar involvement.⁵

The purpose of this report is to describe the perioperative anesthetic management of 3 children with CCAM who underwent lobectomy.

CASE SERIES

Case 1

A 2-year-old female weighing 10 kg presented at the emergency room in mild respiratory distress with SpO₂ on room air ranging between 95-99%. Chest x-ray (Figure 1A) and CT scan showed a congenital cystic adenomatoid malformation measuring 10.8 x 9.4 x 7.8 cm in the right upper lobe.

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In the operating room, standard monitors were attached. The patient was premedicated with IV midazolam (0.05 mg/kg). A modified rapid sequence induction (RSI) using IV fentanyl (2 mcg/kg), propofol (2 mg/kg), and rocuronium (1.2 mg/kg) with minimum positive pressure ventilation (PPV) was performed. Because of the unavailability of an appropriately sized double-lumen tube or bronchial blocker in the institution, the airway was secured using a single-lumen cuffed endotracheal tube (ETT) size 4.5. An arterial line was inserted at the left femoral artery, and a central line was inserted at the right internal jugular vein. An intercostal nerve block (ICNB) using bupivacaine 0.25% was injected at two dermatomal segments above and below the incision site. Anesthesia was maintained with sevoflurane 2-3 vol% and muscle relaxation with 0.3 mg/kg of rocuronium.

Lung protective strategies (TV 6 mL/kg, PEEP 4 cm H₂O) were utilized with two-lung ventilation and continuous ET-CO₂ monitoring. Retractors were used to facilitate surgical exposure of the operative field. There was an increase in airway pressures during retraction and lobectomy, which resolved spontaneously after manipulation. The patient was hemodynamically stable throughout the 6-hour surgery without any inotropic support. Total blood loss was 200 mL which required a blood transfusion. At the end of the surgery, blood clots and secretions in the airway were removed through chest clapping and ETT suctioning before extubation. Point of care testing revealed normal acid-base balance and low airway pressures. Neuromuscular blockade was fully reversed with Sugammadex 8 mg/kg. The patient tolerated on-table extubation, and ventilation was supported using non-invasive positive pressure ventilation (NIPPV) with low airway pressure support. The postoperative analgesia regimen given were IV ketorolac (0.5 mg/kg/dose), paracetamol (10 mg/kg/dose) and nalbuphine (0.1 mg/kg/dose) to supplement the ICNB.

At the post-anesthesia care unit, rhonchi were noted on auscultation, and a postoperative chest radiograph revealed atelectasis on the right lower lung field (Figure 1B). Airway and alveolar issues were relieved with epinephrine (2.5 mL

from 1 mL 1:1,000 L-epinephrine + 4 mL normal saline solution), nebulization, IV dexamethasone (0.5 mg/kg/dose), and judicious chest physiotherapy. The patient was then transferred to the ICU for monitoring.

In the succeeding days, the patient had a FLACC score of 4-6, partially relieved with rescue opioid medications. A repeat chest x-ray showed better aeration in the remaining lung fields (Figure 1C). However, the patient developed hospital-acquired pneumonia, which necessitated antibiotic therapy. The patient improved and was discharged 17 days after.

Case 2

The patient is a 6-month-old 6.5 kg male. He was born to a 40-year-old G₃P₃ mother who tested positive for COVID-19. A patient's chest x-ray showed cystic foci, initially considered part of the infectious process. The patient was discharged well and stable. In the interim, the patient developed dyspnea and poor activity. He was subsequently admitted to the hospital for community-acquired pneumonia. Chest x-ray (Figure 2A) and chest CT scan confirmed an air-filled cystic foci about 2.3 x 2.4 x 1.7 cm and 2.3 x 1.7 x 1.3 cm on the right lower lobe. Mediastinal structures remained in the midline.

In the operating room, standard monitors were attached to the patient. A modified RSI using IV fentanyl (2 mcg/kg), ketamine (3 mg/kg) and rocuronium (1.2 mg/kg) with PPV was performed. The patient was intubated using a single-lumen uncuffed endotracheal tube (ETT) size 3.5. Before cutting, an ICNB was done. The arterial line and central line were inserted. Anesthesia was maintained with sevoflurane at 2-3 vol% while muscle relaxation was achieved with rocuronium. Surgical exposure of the operative field was adequate with retraction. Two-lung ventilation was maintained with lung protective strategies (TV 6mL/kg, PEEP 4 cm H₂O).

The surgery lasted 5 hours with an estimated blood loss of 150 mL, necessitating blood replacement. At the end of the surgery, chest physiotherapy and ETT suctioning were



Figure 1. Chest x-ray of a 2-year-old female with CCAM at the right lobe. (A) Preoperative chest x-ray showing a large cystic structure in the right upper lobe. (B) Postoperative chest radiograph day 0. (C) Postoperative chest radiograph day 3.

done to remove blood clots and mucous secretions along the airway.

With a normal ABG and acceptable low airway pressures, the patient tolerated on-table extubation to NIPPV. The postoperative analgesia regimen were IV paracetamol (10 mg/kg/dose), ketorolac (0.5 mg/kg/dose), nalbuphine (0.1 mg/kg/dose) to supplement the ICNB.

Post-op chest x-ray showed atelectasis on the left lung field (Figure 2B). This was managed with prudent chest physiotherapy, epinephrine (2.5 mL from 1 mL 1:1,000 L-epinephrine + 4 mL normal saline solution) nebulization and IV dexamethasone (0.5 mg/kg/dose). A repeat chest x-ray the following day showed increased aeration at the left lung, atelectasis on the right middle lobe, and significant pleural effusion. A chest tube was subsequently inserted. The patient alternated between left lateral decubitus and prone positions to maximize oxygenation. He had a FLACC score of 4-5, partially relieved with rescue doses of opioids.

With the persistence of right lung atelectasis, the surgical team decided to reintubate the patient and check for obstruction in the airway under fiberoptic bronchoscopy. Intraoperative findings showed copious mucous secretions

and an inflamed right middle lobe bronchus. The bronchial stump at the lower lobe was intact. Given these findings, hypertonic saline nebulization was alternated with salbutamol and n-acetylcysteine nebulization. Judicious chest physiotherapy and regular ETT suctioning were also done. Intravenous dexamethasone was continued for a few more days. OGT was inserted to facilitate feeding. Sedation was maintained with dexmedetomidine (0.5-0.7 mcg/kg/hr) while on low airway pressure support.

Chest x-ray the following morning showed regression of atelectasis on the right middle lobe with better aeration. However, beginning infiltrates were noted on the left lung field (Figure 2C). The patient was treated for nosocomial pneumonia and was discharged after completing ten days of antibiotic therapy.

Case 3

The patient is a 1-year-old male weighing 9.2 kg. He was born full term to a then 30-year-old G₃P₃ mother. The patient presented with a 3-month history of cyanotic episodes associated with fits of cough and colds. SpO₂ on room air was at 99%. Chest x-ray (Figure 3A) and chest CT scan

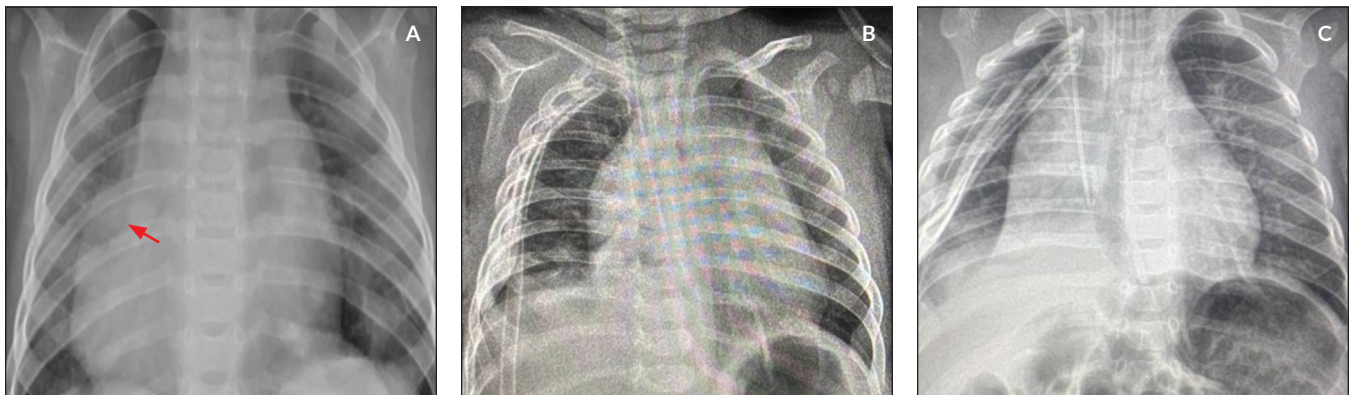


Figure 2. Chest x-ray of a 6-month-old male with CCAM at the right lobe. (A) Preoperative chest radiograph showing multi-cystic lesions at the right lower lobe. (B) Postoperative chest radiograph day 0. (C) Postoperative chest radiograph day 1 (reintubated).

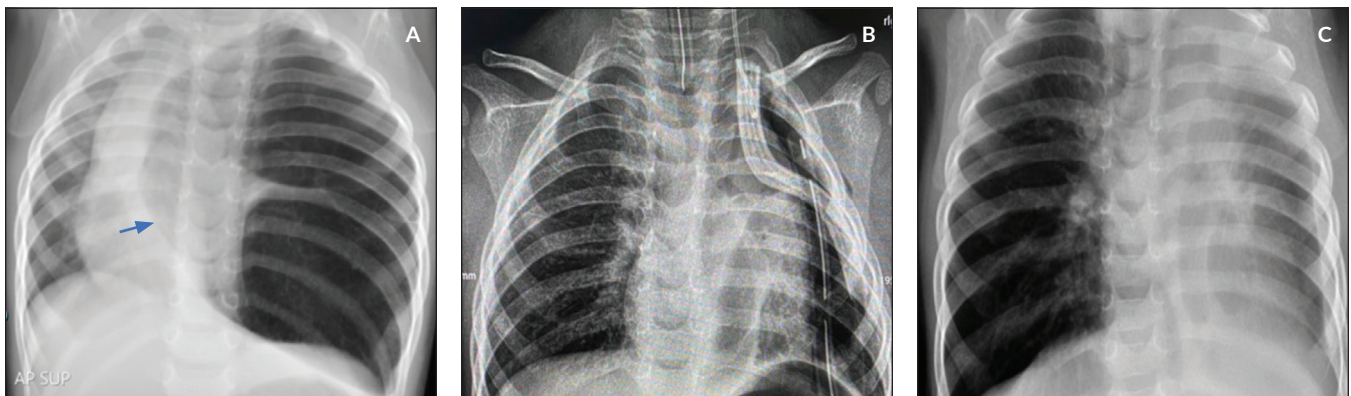


Figure 3. Chest imaging of a 1-year-old male with CCAM at the left lobe. (A) Preoperative chest x-ray showing a cystic left upper lobe with hypoplastic lingula. (B) Postoperative chest radiograph within 6 hours post-surgery (intubated). (C) Postoperative chest radiograph day 3.

Table 1. Patient's clinical profile, surgical details, anesthetic management, and postoperative course

	Case 1	Case 2	Case 3
Age/Sex	2 yr/F	6 mo/M	1 yr/M
Weight (kg)	10	6.5	9.2
ASA status	3	2	2
Diagnosis	CCAM type 1 ^a	CCAM type 1	CCAM type 1
Surgery	Axillary thoracotomy, right upper lobectomy	Right posterolateral thoracotomy, right lower lobectomy	Axillary thoracotomy, left upper lobectomy, postoperative bronchoscopy
Anesthetic plan	GETA + ICNB	GETA + ICNB	GETA + CTEA
Operative time	5 hours	5 hours	3 hours
Estimated blood loss	200 mL	150 mL	20 mL
Blood transfusion	pRBC 100 mL	pRBC 100 mL	None
Intraoperative findings	All right lobes encased in dense adhesion, large cystic lesions located in the right upper lobe, mostly in the anterior segment, viable middle and lower lobes	2 cm cystic structures at the right lower lobe, dense adhesions	Cystic left upper lobe 13.4 x 7 x 4 cm, hypoplastic lingula
Laterality	Right	Right	Left
Two-lung ventilation	Yes	Yes	Yes
Pain regimen	IV: paracetamol, ketorolac PNB: ICNB 0.25% bupivacaine	IV: paracetamol, ketorolac, nalbuphine PNB: ICNB 0.25% bupivacaine	IV: paracetamol, ketorolac Neuraxial: CTEA 0.25% bupivacaine infusion
Mechanical ventilatory support	Immediately extubated to NIPPV	Immediately extubated to NIPPV	Maintained intubation for 6 hours post-resection. Extubated to low flow nasal cannula
FLACC score (day 0 - 3)	4 - 6	3 - 4	0

CCAM - congenital cystic adenomatoid malformation; GETA - general endotracheal anesthesia; ICNB - intercostal nerve block; CTEA - continuous thoracic epidural anesthesia; pRBC - packed red blood cells; PNB - peripheral nerve block; NIPPV - non-invasive positive pressure ventilation

^a CCAM that measures 2 to 10 cm in diameter

revealed a left cystic lucency about 6.3 x 4.6 x 9.3 cm with a rightward deviation of mediastinal structure and compressed left lower lobe.

In the operating room, standard monitors were attached. The patient was premedicated with midazolam (0.05 mg/kg). The induction of anesthesia was carried out using modified RSI with IV fentanyl (2 mcg/kg), propofol (2 mg/kg), and rocuronium (1.2 mg/kg). The airway was secured using a single-lumen uncuffed endotracheal tube (ETT) size 4.0. Lung protection strategies were used, and the respiratory rate was set to maintain normal $ETCO_2$. The arterial line and central line were inserted. Thoracic epidural anesthesia (TEA) was performed using a midline approach at T7. Anesthesia was maintained using combined general anesthesia and epidural technique with sevoflurane at 2 vol% and intermittent boluses of 0.125% bupivacaine at 1 mL/kg/dose, respectively. The surgery lasted 3 hours with an estimated blood loss of 20 mL. The patient remained hemodynamically stable with no episodes of desaturation.

Postoperatively, the patient was kept intubated with mechanical ventilatory support set at low airway pressures to keep the conductive airway patent and prevent atelectasis from airway obstruction due to inflamed bronchus and retained blood clots or mucous secretions. Airway and alveolar issues were ameliorated by giving epinephrine (2.5

mL from 1 mL 1:1,000 L-epinephrine + 4 mL normal saline solution) nebulization and IV dexamethasone (0.5 mg/kg/dose). Chest x-ray showed aerated remaining lung fields with an air bronchogram (Figure 3B).

The patient was extubated uneventfully 6 hours post-surgery. He had no signs of respiratory distress and tolerated low-flow oxygen support via nasal cannula. Chest x-ray (Figure 3C) showed aerated lung fields. Analgesic regimen included an epidural infusion of 0.25% bupivacaine (0.2 mg/kg/hr), IV paracetamol (10mg/kg/dose) and ketorolac (0.5 mg/kg/dose).

The patient was transferred to the ICU for monitoring. He had adequate pain control with a FLACC score of 0. He was subsequently discharged after two weeks. No other postoperative complications occurred during the hospital stay.

Table 1 summarizes the patient's clinical profile, surgical details, anesthetic management, and postoperative course.

DISCUSSION

Among congenital lung anomalies, CCAM ranks second to congenital lung emphysema. Patients may present with other concomitant congenital anomalies.⁶ No other co-existing congenital anomalies were identified in these three patients. In terms of lobe involvement, the left upper lobe,

right middle lobe, and right upper lobe were equally affected. This is slightly different from existing literature stating 41%, 34%, and 21 % involvement of the left upper lobe, right middle lobe, and right upper lobe, respectively.⁶

The perioperative challenges in anesthesia for thoracic surgery are maintaining hemodynamic stability and adequate oxygenation in pediatric patients with open chests placed in lateral decubitus position. General anesthesia is favored because it provides control of the airway and ventilation, steady hemodynamics, immobile patient, and balanced anesthesia. Anesthetic management includes tracheal intubation, controlled ventilation, muscle relaxation, maintenance with an inhalational agent, and adequate analgesia. Intraoperative procedural concerns are lateral position, difficult access to the patient by anesthesiologist post-draping, ventilation, and oxygenation, ETT movement, hemodynamic status, mechanical arrhythmias, bleeding, and cross lung contamination. Monitoring should include standard ASA monitors with an arterial catheter and central line if extensive blood loss, frequent blood sampling, and infusion of vasoactive drugs are expected.⁶

Premedication with intravenous midazolam was given to decrease the likelihood of agitation and crying as crying-induced forceful inspiratory efforts may lead to air trapping in the cyst.⁶ Likewise, intravenous induction was chosen over inhalational induction since the conduct of inhalation induction may elicit brief crying episodes. Reduced uptake of inhalational agents can delay inhalational induction due to intrapulmonary shunting in patients with lung pathology.⁶ In a report by Takroui et al., inhalation induction was prolonged and led to an inadequate intubating condition requiring rescue administration of ketamine and rocuronium to secure the airway.⁷ Modified rapid sequence induction was conducted as paralysis facilitates intubation and ensures low peak airway pressures.⁶ Also, this allowed maximizing the oxygen reserves of the patients to reduce the likelihood of rapid desaturation that occurs with classical RSI.⁸

Conventional two-lung mechanical ventilation is known to cause expansion of the nearby lung lobes due to the enlargement of cysts by ball-valve air entrapment. Cyst resection may require one-lung ventilation (OLV) as it provides better surgical access, causes less bleeding, and gives protection from contralateral lung contaminants.⁶ This is technically more challenging to perform in young patients. In addition, pediatric OLV devices such as pediatric double-lumen tubes, uninvent tubes, and bronchial blockers may not be readily available in every institution.

In the patients presented, traditional single-lumen ETT appropriate for age and weight was used due to the unavailability of equipment. Since these were open thoracotomies, surgical exposure of the operative field was accomplished through mechanical retraction. These retractors were intermittently released when airway pressures were elevated as high peak airway pressures during positive pressure ventilation increases the risk for barotrauma. Successful

lobectomy and pneumonectomy via open thoracotomy in infants with CCAM using single lumen ETT and manual retraction have similarly been reported.^{9,10}

Ensuring adequate gas exchange while avoiding atelectasis and iatrogenic barotrauma were issues of concern for these patients. Infants and young children have smaller functional residual capacity (FRC), larger closing volume, and greater chest wall compliance making them more susceptible to atelectasis during general anesthesia.¹¹ Unlike adults, wherein V/Q matching is improved with lateral decubitus position, i.e., oxygenation is optimal when the healthy lung is dependent (“down”) and the diseased lung is non-dependent (“up”), the opposite effect is observed in infants wherein oxygenation is improved with the healthy lung “up” and the diseased lung “down”.^{12,13} This is primarily due to the soft compliant ribcage of infants.

In a single-center study by Lee et al. assessing the effects of ventilatory strategies in pediatric thoracic surgery, they observed fewer pulmonary complications within three days in patients who received lung protective strategy (TV at 6 mL/kg during two-lung ventilation) compared to those who received conventional mode of ventilation (TV at 10 mL/kg).¹¹

CCAM may contain fluid. Lung protective strategies reduce the risk of spillage. In addition, anesthesiologists must be vigilant with ETT suctioning before and after a shift in patient position to prevent lung contamination.

Ensuring adequate postoperative pain control is vital to reduce intraoperative anesthetic requirements, promote early extubation and prevent postoperative splinting. A case report by Fajardo-Escolar et al. showed that the infant who received continuous epidural infusion was eligible for extubation immediately after the surgery.¹⁴ ICBN is effective as well.⁹ In the patients presented, continuous epidural analgesia using bupivacaine afforded better analgesia than single-shot ICNB. It is possible that the continuous nature of the epidural analgesia in the patients presented was the factor that led to better pain control.

Spinal cord injury after inserting a thoracic epidural catheter during general anesthesia emphasizes the importance of weighing the risk-benefit ratio of placing an epidural catheter on the thoracic area.¹⁵ This should be reserved for extended thoracic procedures and must be performed by experienced anesthesiologists. Alternatively, an epidural catheter can be introduced in the caudal space and threaded cephalad to the thoracic level.^{9,10} Ultrasound guidance further increases the safety of its performance.

Neonates and infants are more sensitive to opioids. Hence, there must be caution when opioids are administered. Small children are more prone to apnea from the imbalance of mu-receptors and increased susceptibility to hypoventilation because of the decreased ventilatory response to hypoxia and hypercapnia. Nalbuphine was chosen as activation of the kappa receptors causes analgesia without significant respiratory depression.¹⁶

Pediatric patients can tolerate on-table extubation provided that children have a relatively good preoperative condition, uneventful intraoperative course, low airway pressures, replaced blood loss, normal acid-base balance, and fulfilled criteria for extubation. Early extubation prevents iatrogenic injury-induced bronchial stump dehiscence from PPV support.⁶ Despite lung protective strategies, intraoperative double lung ventilation most likely compressed the healthy lung. Thus, there should be a high index of suspicion for possible postoperative atelectasis.

If extubation is attempted in a patient with risk for laryngeal edema post-thoracic surgery, a nebulized commercially available racemic epinephrine nebulization (1:1 mixture of Dextro D and Levo L isomers of epinephrine) or L-epinephrine nebulization can improve the inflammatory process in the airway by the reduction in respiratory secretions and mucosal edema (alpha-adrenergic effects) and relaxation of the airway smooth muscle. L-epinephrine is less expensive and more widely available. Nebulization should not be given more frequently than every 2 hours, and one should be mindful of the potential side effects such as tachycardia, hypertension, arrhythmia, and pallor.¹⁷ Studies comparing racemic epinephrine and L-epinephrine nebulization suggest that both equally effectively treat post-extubation laryngeal edema.¹⁷ Although limited data is available, dexamethasone (0.5mg/kg/dose) administered 6-12 hours prior to extubation, then every 6 hours for six doses is used for pediatric patients with airway edema prior to extubation.¹⁸

The postoperative course depends on the surgical procedure and underlying disease. A systematic review and meta-analysis of the postnatal management of congenital cystic lung lesions reported early postoperative complications such as air leak, pleural effusion, pneumonia, atelectasis, bleeding, wound infection, and respiratory failure. Late complications include asthma, recurrent pneumonia, and residual disease.¹⁹

Even though it was the 3rd patient who presented with preoperative mediastinal shift on imaging, he did not develop atelectasis and pneumonia postoperatively, unlike the 1st two patients. The timing of extubation and analgesic regimen most likely contributed to this. Since atelectasis was anticipated based on the experience with the two previous patients and the preoperative finding of mediastinal shift and hypoplastic lingula, the team kept the patient intubated post-surgery and allowed the lungs to expand adequately before extubation. Concomitant provision of continuous neuraxial analgesia provided better analgesia based on observed pain scores leading to a smoother recovery.

CONCLUSION

In children with CCAM for open thoracotomy, modified RSI can be used safely among those without difficult airway features and significant cardiorespiratory compromise. In limited resource settings, intraoperative double lung venti-

lation with lung protective strategies (TV 6 mL/kg, PEEP 4 cm H₂O) and postoperative use of dilute L-epinephrine for nebulization produce satisfactory results. Continuous epidural analgesia appears to provide better pain control compared to single-shot ICNB. Preoperative conditions and intraoperative course are significant determinants in the timing of extubation. Whenever permissible, early extubation is sought to avoid iatrogenic injuries. Atelectasis due to airway edema and mucus/blood clots along the conductive airway are common postoperative issues that should be aggressively monitored and treated.

Declaration of Patient Consent

The authors certify that they have acquired appropriate consent forms from the parents of the patients granting usage of clinical information and diagnostic images for publication. They understand that the names will not be published, and due efforts were made to conceal the identity.

Statement of Authorship

Dr. Ralph Philip M. Anislag. The author contributed to writing both the original and final manuscript. Dr. Grace G. Catalan. Both authors reviewed and approved the final submitted case series.

Author Disclosure

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