Review Article

Anesthetic considerations in patients with cystic pulmonary adenomatoid malformations

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Abstract

Congenital pulmonary adenomatoid malformation (CPAM) is a rare entity. The authors searched the US National Library of Medicine Database, EMBASE, Google Scholar, PubMed Central for anesthetic management in CPAM. The search was performed using the terms: congenital cystic adenomatoid malformation, congenital pulmonary adenomatoid malformation, CCAM, CPAM, anesthetic management. The prognosis of CPAM depends on timely diagnosis, presence of hydrops, degree of hypoplasia of remaining lung, and the size of the lesion. Symptomatic patients must be treated surgically and lobectomy is considered the gold standard. Anesthetic management of such cases is challenging as it involves thoracotomy or thoracoscopic lobectomy or cystectomy and can lead to sudden hemodynamic Collapse. Early extubation should be considered to avoid iatrogenic ventilator-induced bronchial stump dehiscence resulting from positive pressure ventilation.

Keywords: Airway concerns, anesthesia management, complications, congenital cystic adenomatoid malformation (CCAM), congenital pulmonary adenomatoid malformation (CPAM), one-lung ventilation

Introduction

Congenital pulmonary adenomatoid malformation (CPAM), earlier known as congenital cystic adenomatoid malformation (CCAM), is a rare bronchopulmonary congenital anomaly characterized by a mass consisting of excessive proliferation of bronchi, but without normal alveolar development. CPAM usually requires surgery in the form of cystectomy or lobectomy or thoracotomy and excision of lesions. Anesthetic induction can lead to sudden hemodynamic collapse. Moreover, positive pressure ventilation (PPV) may cause respiratory embarrassment.[1-3] The review discusses the anesthetic considerations and optimal management.

Literature Search

Keeping these words ("congenital cystic adenomatoid malformation," "congenital pulmonary adenomatoid malformation," "CCAM," "CPAM," "anesthetic management") as the main keys either in single or in various combination, an online search was performed using US National Library of Medicine Database, EMBASE, Google Scholar, and PubMed Central without any language restriction or limits with keywords, from the years 1949 till 31st July 2020. It revealed 4,000 results; articles of relevance about anesthetic management included 245 searches, with available full texts in 197 searches. The references of relevant articles were cross-checked and the review was imbibed from the articles

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which elaborated on these keywords. Thirty relevant articles, after excluding articles with the duplicity of information and ensuring recent data have been included in this review.

Incidence

CPAM has an incidence of 1/20,000–1/30,000 live births. It accounts for 25% of all congenital lung anomalies, second only to congenital lung emphysema.[1] CPAM may be associated with various other congenital anomalies/malignancies as summarized in Table 1.

The most commonly involved lobe is the left upper lobe in 41% cases, in 34% cases the right middle lobe is involved and in 21% cases there is right upper lobe involvement.[2]

Diagnosis

CPAM may be identified in the fetus in pregnant women who develop polyhydramnios by the 23rd week of gestation. Diagnosis can be made during ultrasound (USG) by the presence of fluid-filled distended lungs.

Chest X-rayfindings may reveal air-filled cysts in the lungs or a solid intrathoracic mass. Large lesions may cause a mass effect with a shift of the mediastinum to the opposite side and depression/inversion of the diaphragm. A computed tomography scan (CT scan) may help to reach a conclusive diagnosis. Real-time assessment remains integral to the appropriate diagnosis.[4-6]

Clinical Course

The natural history of CPAM ranges from spontaneous regression to neonatal death. Up to 1/3rd of cases identified in utero resolve before birth. Small CPAM lesions may remain asymptomatic and get diagnosed incidentally later in life. Larger CPAM lesions are associated with fetal hydrops and severe respiratory failure at birth. In most cases, CPAM manifests as neonatal acute respiratory distress syndrome with or without infection.[3]

Table 1: Associated congenital anomalies/malignancies with CPAM[1-3]

Anomalies	Prevalence
Renal, bony, intestinal and cardiac anomalies	25%
Congenital cardiac disease, pectus excavatum pulmonary and vascular malformations	10%
Associated malignancy pulmonary blastomas bronchial and bronchoalveolar carcinoma rhabdomyosarcoma adenocarcinoma	4-9%

Classification of CPAM

CPAM is classified by the Stocker classification, according to the size of the lung cyst into 3 types, Type I (1–10cm), Type II (0.5–1cm), and Type III (<0.5cm, micro-cystic and grossly solid).[3] CPAM lesions have also been classified by Bale PM, based on clinical presentation and pathological picture into cystic, intermediate, and solid types, with solid having the worst prognosis.[5]

Prognosis and Survival

Prognosis and survival depend on the presence of hydrops, size of the lesion, timely diagnosis, and degree of hypoplasia of the remaining lung.[7] Predominantly solid lesions though often small in size are associated with polyhydramnios, ascites, and fetal anasarca and have a poor prognosis.

Ultrasound diagnosis of CPAM in the fetus should ideally be made as early as possible to allow timely intervention especially in the case of hydrops.[8] The measurement of the CPAM volume ratio (CVR) normalized for gestational age has been observed to be the best indicator to predict outcomes. CVR is defined as CPAM volume divided by head circumference (measured in cm). CPAM volume is measured by the formula: Length × Height × Width × 0.52. The CVR can be calculated by the formula: Length × Height × Width × 0.52)/ head circumference. A CVR greater than 1.6 predicts the development of hydrops.[8]

Differential diagnosis of fetal lung masses[6]

1. Congenital pulmonary malformations
2. Congenital diaphragmatic hernia- Bochdalek, Morgagni, Para-esophageal hernia
3. Tumors: Neuroblastomas, teratoma, thymic tumor, pulmonary blastomas.
4. Vascular malformation cyst
5. Esophageal duplication cyst.

Management of a fetus with a lung mass

The indications of interventions in the fetus diagnosed with CPAM include hydrops, large lesions, and growing lesions. Serial needle aspirations or ultrasound-guided placement of thoracic-amniotic shunt may be indicated in cases of large solitary lung cysts in the fetus. Fetus CPAM resection is advocated in those in whom thoracocentesis or shunt placement is not possible. Ex-Utero intrapartum therapy (EXIT) procedure is indicated when the diagnosis is delayed and

the lesion is likely to adversely affect neonatal resuscitation. Peranteau WH *et al.* observed improved survival benefit with steroid administration to the mother if the fetus has hydrops and a CVR >1.6.[9]

Management of a neonate with a lung mass

An asymptomatic stable neonate can be managed without active intervention.[10]

Symptomatic patients must be treated surgically and lobectomy is considered the gold standard. Segmentectomy and wedge resection are performed when feasible and observed to be as efficacious as lobectomy; the remaining unaffected lung should be preserved. After complete CPAM excision, the prognosis is usually good.[6]

Timing of surgery for CPAM

This is controversial and emergent, urgent or elective surgery is determined by prenatal behaviour, development of mass effects, and postnatal presentation.[11] Table 2 shows the summarized indications and presentations for the timing of surgery.

Improved postoperative outcomes have been observed in children with small lesions undergoing elective surgery when they are asymptomatic compared to those undergoing surgery only after the development of symptoms.[12]

Pre-operative Evaluation

A detailed history and clinical examination are essential. Important points to consider in history are summarized in Table 3.The clinical examination may reveal signs of tachypnoea, reduced breath sounds, poor oxygen saturation, chest in-drawing, respiratory distress, and abnormal hemodynamic parameters. Investigations indicated before surgery along with their relevance is compiled in Table 3.[13-15] Unstable patients may require preoperative optimization and stabilization in elective cases. Delaying of surgery may be warranted in cases of pneumonia or recurrent chest infection.[13]

Anesthesia for CPAM Surgery

Anaesthesia for Fetal CPAM surgery

Fetal in utero lobectomy is performed in the mid-gestation period and the fetus is then returned into the uterus for further development to term. The first open fetal surgery successfully removed the CCAM lesion and salvaged the fetus.[16] The mother was preoperatively administered indomethacin and nafcillin. Under general anesthesia, the aim was to preserve uteroplacental blood flow by maintaining uterine relaxation with the help of a deep plane of anesthesia (inhalational anesthetic agents) and maintaining uterine volume using amnio-infusion. Abdominal paracentesis was required to facilitate exposure of the right chest for fetal thoracotomy. Intraoperative fetal monitoring was provided by electrocardiography and neonatal radial artery pulse oximeter. Following a right fifth intercostal space thoracotomy, the large middle lobe CCAM was resected. The fetal thoracotomy was closed and the fetus was returned inside the uterus; repeat nafcillin 500 mg was instilled in the amniotic cavity followed by the closure of uterine layers.

EXIT procedures of fetal thoracotomy and lobectomy are usually undertaken in a fetus with large malformations, severely compromised airway, or hydrops and performed before the umbilical cord is clamped. Fetal complications are known to occur during EXIT procedure; causes can be because of cord compression, placental abruption, or loss of uterine relaxation. The neonate may later be connected to an extracorporeal membrane oxygenator (ECMO).[17]

Anaesthesia for Postnatal CPAM Surgery

General anesthesia is preferred, as it provides complete

Table 2: Indications of surgery

Type of surgery	Indication	Presentation
Emergency thoracotomy (within hours)	Neonate in severe distress or with progressive decompensation despite intervention and unlikely to improve by non-surgical management.	Neonate with severe respiratory distress (tachypnea, increased work of breathing, hypoxemia, hypercapnia or overt respiratory failure requiring ventilator support), hypotension, and mediastinal shift
Urgent surgery (within days)	Symptomatic neonate in respiratory distress who is stable after intervention without expected further improvement and with risk of deterioration. Symptomatic infants and young children diagnosed postnatally	Infants and young children with borderline symptoms who become symptomatic or persist with tachypnea, feeding difficulties, and failure to thrive.
Elective surgery (within days to months)	Asymptomatic lesions in infants with small lesions	Stable neonate and infant with moderate symptoms and no distress

Table 3: Preoperative evaluation

Evaluation method	Findings and techniques of evaluation
History	*Newborn and early infancy* Nasal flaring, tachypnea, and intercostal retraction tracheal, airway or mediastinal compressive symptoms *Children and adults* Recurrent history of cough, chest pain, dyspnea on exertion History of recurrent unresolving or resolving pneumonia or pneumothorax and sometimes respiratory compromise
Laboratory Investigations	*Complete blood count*: to know hemoglobin, infection *Serum electrolytes*: depending on clinical condition *Coagulation studies*: in patients presenting with infection or sepsis *Arterial blood gas analysis*: in patients presenting in respiratory distress to know the pO_2, pCO_2 levels and adequacy of ventilation
Radiological Imaging	*Chest x-ray*: to evaluate airway, mediastinal shift, infection etc *CT chest*: for confirmation of diagnosis, location of lesion, solid or cystic masses, infection, airway communication, pulmonary sequestration, associated cardiac defects, mediastinal shift or compression. *MRI*: to know lobe location, identify lesions and status of normal lung. *Echocardiogram*: to assess for congenital associated anomalies of cardiac or respiratory systemand to know cardiac function.
Fetal Imaging (for EXIT procedure)	*Prenatal ultrasound*: to assess lung size, CVR ratio, mediastinal shift, polyhydramnios and fetal hydrops. *Prenatal Doppler*: to assess and identify feeding vessels *Fetal MRI, fetal ultrasonography*: to image suspected lesions, presence of mass, delineate blood supply and to assess residual lung mass. *Fetal echocardiography*: to evaluate cardiac status
Blood group type and cross match	Mandatory as the procedure involves proximity to great vessels and many of the neonates have already impaired cardio-pulmonary function.

control of airway, breathing, and circulation. It provides good operating conditions for the surgery, controlled hemodynamics, an immobile patient, and analgesia and amnesia as desired. Anesthetic management includes tracheal intubation, controlled ventilation, muscle relaxation, maintenance with an inhalational agent in air/oxygen mixture, and adequate analgesia. Intraoperative concerns have been summarized in Table 4.

Conduct of Anaesthesia

Premedication with oral or intravenous midazolam may be required in the older infant with minimal cardio-respiratory compromise. Glycopyrrolate may be given to dry secretions and to prevent vagal mediated bradycardia during laryngoscopy and intubation in neonates.

Monitoring should include standard ASA monitoring and a stethoscope should be placed in all cases on the non-operating side. An arterial catheter and femoral central venous line may be considered if extensive blood loss or resection is expected. Chiluveru *et al.* in addition to standard monitors have also used the femoral line for CVP and invasive blood pressure for their patients with associated atrial septal defect (ASD) and severe pulmonary arterial hypertension (PAH).[18]

Inhalational induction may be delayed in these infants owing to the lung pathologyand reduced uptake of inhalation agents. Intravenous induction with minimum positive pressure ventilation or a spontaneously breathing patient is preferred to prevent distension of the lung cysts. Isoflurane in air/oxygen mixture and sevoflurane with air/oxygen mixture has been successfully used without any untoward event.[13,14,18] Swapna *et al*. successfully used sevoflurane and ketamine in the anesthetic management of a 6-month-old female infant, a diagnosed case of ASD, and bidirectional shunt, severe PAH posted for right upper and middle lobectomy.[18] However, Takrouri M *et al*. observed a longer time for induction and inability to achieve adequate intubating conditions with a fighting patient when using sevoflurane and had to supplement intravenous ketamine for successful intubation attempts.[19]

A modified rapid sequence induction should be preferred to secure the airway and avoid air trapping in the cysts during forceful inspiratory efforts in a crying infant. Thiopentone, ketamine, lidocaine, dexmedetomidine may be used to facilitate the placement of tubes without cardiac or respiratory responses. The child should be maintained on the spontaneous mode of ventilation with sevoflurane in air or oxygen mixture with occasional manual assistance with minimal airway pressure until thoracotomy has been done. Nitrous oxide can accumulate in the cysts with air-fluid levels resulting in emphysematous enlargement and should be avoided. However, it can be supplemented after the cyst has been removed. The rate of increase in inhalational anesthetics levels may be slowed in the presence of intrapulmonary shunting. Muscle relaxation with atracurium or rocuronium can be provided after the chest and pleura are opened. Care should be taken to ensure that low peak airway pressures are maintained.Analgesia may be provided using short-acting opioid analgesics. Guruswamy V

Table 4: Intraoperative Concerns

Procedural step	Procedural concern
Positioning of patient	Lateral position (respiratory effects and decrease in functional residual capacity)
Post draping by surgeons	Difficult access to patient for anesthesiologist
Access to CCAM by surgeon retraction of lungs	Additional ventilation concerns Additional oxygenation concerns Tracheobronchial kinking Tube movement Hemodynamic effects Impaired venous return Aortic compression Mechanical arrhythmias Bleeding
Change in position from lateral to supine	Cross lung contamination

et al. used remifentanil for MAC sparing effects and avoided nitrous oxide considering the risk of cyst expansion.[14]

A Jackson Rees modification of Ayer's T-piece circuit should be available to check chest compliance at various time points during surgery. Bag compliance correlates well with an increase in resistance to retraction of lungs during surgery. Surgeons may be requested to release lung retraction in case the bag is too tight or ventilation is difficult.

Perioperative Ventilation strategies

Resection of the cyst and involved lung lobe may necessitate one-lung ventilation (OLV). Conventional mechanical ventilation is known to cause expansion of the affected lung lobes due to the enlargement of cysts by ball-valve air entrapment. High peak airway pressures may also increase the risk of barotrauma. OLV is desirable to improve the surgical field, minimize trauma to the remaining healthy lung, reduce blood loss, and also reduce contamination of the healthy lung with secretion/spillage from the operative site.

Various types of equipment are available such as pediatric double-lumen tubes (DLT), uninvent tubes, and bronchial blockers (BB) to provide OLV. However, these are not available in sizes suitable for neonates and infants. The smallest available DLT is 26 F, useful only in children above 8 years and the smallest Univent®tube is 3.5 mm ID which can be used in children aged older than 6 years. Guruswamy V *et al.* reported the use of a 3F Fogarty catheter as a BB placed parallel to the endotracheal tube (ETT).[14] Alternatives include a balloon-tipped angiography catheter or a5F Cook pediatric BB. Both can be passed into the required bronchus using a guidewire and have the advantage of an end hole which allows suction and collapse of the lung.[20,21] Bronchial blockers are an effective strategy of lung isolation in small children; they can be introduced alongside a conventional ETT and positioned at a desirable level using a fiber optic bronchoscope (FOB). A 5F bronchial blocker is a better option compared to a Fogarty catheter which may cause mucosal damage because of the high-pressure balloon.[18] The embolectomy catheter balloon should be inflated with small incremental volumes of air, till the seal is achieved and there is no air leak. Correct positioning of the BB should be ascertained through a repeat FOB check. The risk of hypercarbia during inflation of the BB needs special mention.[22]

The other desired approach is to use a conventional ETT and to advance it into the desired bronchus with the help of a FOB. This may not, however, provide a completely collapsed lung. The disadvantages include difficulty in suctioning of secretions of the non-ventilated lung and obstruction of the upper lobe bronchus. An increase in dead space is also a cause of hypoxia in such cases. A drop in saturation after instituting OLV with this technique has been reported by Chiluveru *et al*.; the saturation could not be improved despite bringing back the ETT into the trachea. They observed improvement in saturation only on decompression of lungs by inserting an 18G cannula in the right 2nd intercostal space, after which surgery could proceed once re-positioning of the ETT into left main bronchus was achieved.[18]

The need for OLV has been questioned.Some surgeons may not request for collapsing the lung, as they retract the lung, providing adequate exposure at the operative site.[21,22] The decision of OLV should be discussed with the surgeon beforehand, as the provision of OLV requires an experienced anaesthesiologist.[23]

High-frequency oscillatory ventilation may be preferred in neonates with compromised respiration as it helps lowers peak airway pressure and the possibility of barotrauma and prevents enlargement of air spaces in air-filled cysts.[13, 15]

Lee *et al.* assessed the effects of perioperative ventilator strategies prospectively in children aged less than 5 years diagnosed with CPAM who underwent VATS lobectomy or segmentectomy in a randomized controlled trial. They

observed less pulmonary complications within 72 h when using lung-protective ventilation (tidal volume at 6 mL/kg during two lung ventilation and 4 mL/kg during OLV, with 6 cmH_20 PEEP) when compared with conventional mode of ventilation (tidal volume at 10 mL/kg and 8 mL/kg during two lung and OLV, respectively, without PEEP).[21]

Positioning concerns

Surgery is usually performed in the lateral decubitus position. This position is known to cause respiratory effects and a decrease in functional residual capacity under anesthesia. Furthermore, there may be dislodgement of theETT. Soft padding in appropriate locations should be done to avoid nerve compression.

Intraoperative spillage concerns

CPAM may contain fluid varying from clear to purulent.[23]The risk of spillage may be reduced by minimal positive ventilation before isolating the non-operative lungandsuctioning at routine intervals in the intraoperative period. One must ensure to suction the ETT in the lateral position, before the patient is made supine, to prevent cross lung contamination at the end of surgery.

Perioperative Regional Anaesthesia Supplementation Techniques

Thoracic epidural catheter placement is performed after induction, to provide both intraoperative and postoperative pain relief, allow for early extubation, and reduce anesthetic requirements. However, one must ensure a normal coagulation profile, and an experienced anesthesiologist to achieve neonatal epidural placement. Bupivacaine or ropivacaine 0.1% with fentanyl 2–3 μg/mL or morphine may be used postoperatively for 48–72 h. There are various reports of the use of epidural or morphine infusionfor postoperative analgesia.[23-28] Studies in children have suggested an epidural technique to be better than morphine infusion.[25] Bosenberg demonstrated a significant beneficial effect from epidural analgesia in 240 neonates.[26] Ultrasound may be reserved for the identification of epidural space in difficult cases. Incorrect positioning of the caudal cannula is common, and therefore ultrasound saline bolus confirms position and rules out intravascular, intrathecal, or intraosseous placement. Guruswamy *et al*. have used a Sono site 180 Plus ultrasound machine to confirmed the presence of epidural cannula by recording the anterior displacement of dura mater following injection of 2ml saline test bolus.[14]

Intercostal nerve block (ICNB) can be provided by the operating surgeon before the closure of the wound or anesthesiologist after completion of the surgery, at two dermatomal segment levels above and below the surgical incision. Gupta L *et al.* reported providing adequate analgesia and early extubation with ICNB infiltration of local anesthetics (LA) in a child with CPAM.[13] The infant had excellent postoperative analgesia and could breathe with good respiratory efforts. However, LA toxicity is a concern as it is known to be rapidly absorbed via intercostal spaces.

Port-site LA infiltration may provide intra and postoperative pain relief with the thoracoscopic approach. Port sites are considered most notorious for causing pain; hence relief should be adequate. Gupta L*et al.* report successful analgesia using port infiltration of LA, in a child who presented with sepsis and long-standing illness associated with CPAM as they could not place a thoracic epidural.[13]

Postoperative Care

The child can be extubated on the table after completion of the surgery, provided the preoperative condition was good, intraoperative course uneventful and standard extubation criteria are met. Early extubation is desirable to avoid iatrogenic ventilator-induced bronchial stump dehiscence by PPV. In neonates who require postoperative ventilation, high-frequency oscillation, and ECMO has been tried. Adequate pain relief and fluid administration should be ensured in the postoperative period.[29] Epidural analgesia should be continued postoperatively as results have suggested early and better recovery with epidural opioids as compared to systemic opioids.[29] The postoperative course depends on the extent of the surgical procedure and underlying diseases.[30] Pulmonary hypertension is one of the major causes of morbidity and mortality in these children.

Postoperative complications

Precipitous hypotension with volatile agents can occur in patients with low cardiac reserve. Respiratory complications reported in a meta-analysis of 9 studies revealed double the number of postoperative complications in symptomatic versus asymptomatic patients (risk ratio 2.8, $P < 0.005$). Complications arising following emergency surgery were noted to be significantly higher (28%) than for elective surgery in neonates and infants.[25] Early pulmonary complications include hypoventilation, desaturation, air leak, pleural effusion, bleeding, and infection. Late complications include residual disease. Serial radiographs and CT scans are required to determine residual disease, depending on the presence/absence of respiratory symptoms during the first 6months until the age of 1 year.

Conclusion

Successful management of congenital cystic pulmonary

adenomatoid malformations (CPAM) in babies needs multi-disciplinary care and anesthesiologists form an important part of the team. Anesthetic management of such cases is challenging as it can lead to sudden hemodynamic collapse and hypoxia during induction. Lung protective ventilation strategies and thoracic epidural catheters are recommended. Early extubation should be considered to avoid iatrogenic ventilator-induced bronchial stump dehiscence. Provision of care by expert anesthesiologists is essential for a successful outcome.

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Conflicts of interest

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