



Experience of video-assisted thoracic surgery treatment of congenital pulmonary airway malformation in infants less than 3 months of age

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Background: Congenital pulmonary airway malformation (CPAM) is the most common type of congenital lung malformation, some children were born with serious respiratory and circulatory disorders, these children tend to have larger cystic lesions and emergency surgery should be taken after birth. This article aimed to explore the feasibility of video-assisted thoracic surgery (VATS) for CPAM in infants less than 3 months of age and analyze the early and mid-term postoperative outcomes.

Methods: A retrospective analysis was performed on 12 children with CPAM who were admitted to Shanghai Children's Hospital from January 2019 to December 2022. Among them, there were ten boys and two girls. The age ranged from 13 to 89 days, with an average of 60.09 ± 30.13 days. The body weight ranged from 2.8 to 7.5 kg, averaging 5.12 ± 1.56 kg. All patients had severe respiratory symptoms such as persistently tachypneic, recurrent pneumonia or respiratory failure.

Results: All of the included children underwent VATS with no cases converted to thoracotomy. The operation time ranged from 40 to 190 minutes, with an average of 124.17 minutes. Among the children in this study, ten cases underwent lobectomy; one underwent segmental resection; one underwent irregular resection. Postoperative mechanical ventilation time ranged from 3 to 49 hours, with an average of 12.75 hours. The indwelling time of the thoracic drainage tube ranged from 2 to 9 days, with an average of 3.92 days. The postoperative hospital length of stay ranged from 5 to 12 days, averaging 8.42 days. None of the children had postoperative complications such as bleeding, bronchopleural fistula or atelectasis. The follow-up time ranged from 3 months to 4 years, and all the children were discharged within 12 days of postoperative hospitalization without need for readmission. Furthermore, no residual lesions were found in the reexamined chest computerized tomography (CT), the lung inflammation dissipated, and no residual cavity was in the affected chest.

Conclusions: Infants with symptomatic CPAM less than 3 months of age should be operated on as soon as possible. With the improvement of minimally invasive technology and anesthesia management, VATS is relatively safe and effective in treating children with such conditions.

Keywords: Congenital pulmonary airway malformation (CPAM); infants; video-assisted thoracic surgery (VATS)

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Introduction

Congenital pulmonary airway malformation (CPAM) is the most common type of congenital lung malformation (CLM), a congenital pulmonary hamartoma-like lesion characterized by excessive proliferation and dilatation of terminal bronchioles (1). Additionally, it often manifests as a single-chamber or multi-chamber cyst or honeycomb structure in lung parenchyma (1). The prevalence of CPAM accounts for about 1/35,000–1/7,200 of the live fetuses, and this rate is gradually increasing (2). For asymptomatic children with CPAM after birth, the appropriate age for operation is above 3 months old (3,4). Notably, a few children were born with serious respiratory and circulatory disorders due to the compression of the heart and lungs and mediastinal deviation. Generally, these children tend to have larger cystic lesions, and emergency treatment should be taken after birth. At present, video-assisted thoracic surgery (VATS) to treat CPAM has gradually become mainstream. However, there are few studies on the use of VATS to treat CPAM in infants of less than 3 months old. Therefore, we collected the case data of CPAM in infants under 3 months of age admitted to Shanghai Children's Hospital and reported on our treatment experience. We present this article in accordance with the STROBE reporting checklist (available at <https://tp.amegroups.com/article/view/10.21037/tp-23-475/rc>).

Highlight box

Key findings

- Video-assisted thoracic surgery (VATS) is relatively safe and effective in treating infants with congenital pulmonary airway malformation (CPAM) accompanied by severe clinical symptoms less than 3 months of age.

What is known and what is new?

- At present, VATS to treat CPAM has gradually become mainstream which has more advantages compared with thoracotomy.
- There are few studies on the use of VATS to treat CPAM in infants of less than 3 months of age. We shared our treatment experience in this article.

What is the implication, and what should change now?

- With the improvement of minimally invasive technology and anesthesia management, VATS is a safe and effective surgical approach. But the learning curve may be longer than that of thoracotomy.

Methods

Clinical data

The relevant data of 12 children with CPAM admitted to our hospital from January 2019 to December 2022 were collected (Table 1). There were 10 males and 2 females. The age ranged from 13 to 89 days, averaging 60.09 ± 30.13 days. Additionally, the body weight ranged from 2.8 to 7.5 kg, averaging 5.12 ± 1.56 kg. All 12 cases were found with lung lesions by ultrasonic examination from 18 to 24 weeks of gestation, and all were born at term. Moreover, the CPAM volume-ratio (CVR) values ranged from 0.68 to 4.12, averaging 1.95 ± 1.03 . As the huge CPAM was discovered in utero, all these children were admitted to the neonatal intensive care unit directly after they were born in the local hospitals. Most of them were born with symptoms such as tachypnea. Due to lack of medical conditions for surgery in those local hospitals, the parents of these children chose our hospital for surgery. Six children were admitted because of tachypnea, accompanied by dyspnea or cyanosis of the mouth and lips. Four cases had a history of repeated cough and fever, chest X-ray suggesting pulmonary inflammation, and the conservative treatment had poor results. One of them suffered from lung consolidation owing to repeated infections in the lower lobe of the right lung. Furthermore, two cases were treated in the intensive care unit of a local hospital due to respiratory failure combined with pulmonary infection, requiring mechanical ventilation. They were subsequently transferred by ambulance to our hospital for treatment. All children were examined by computerized tomography (CT) before operation (please see Figures 1–4 for CT of some cases).

Operative method

All children underwent VATS at Shanghai Children's Hospital. Operative methods were divided into lobectomy, segmentectomy, and lung-sparing resection. The three-dimensional CT imaging was carefully analyzed before the operation to determine the resection range, which may be adjusted according to intraoperative findings during the operation. One-lung ventilation was selected for anesthesia (bronchial occluder was used to selectively occlude the main bronchus of the affected side), a closed thoracic cavity was established, and artificial pneumothorax was maintained (pressure was 3–4 mmHg, flow rate was about 1 L/min) (1 mmHg = 0.133 kPa). For children with giant vesicles, we tried to gain more operating space by cauterizing the giant vesicles. In the lateral position of the child, three-

Table 1 Patient characteristics

Pt No.	Age (d)	Weight (kg)	CVR	Operative method	OPT (min)	MVT (h)	ITOTDT (d)	HST (d)	Stocker type
1	85	7.5	1.79	Lobectomy	135	4	3	9	1
2	89	6.4	0.69	Lobectomy	100	5	3	6	3
3	31	4.6	1.62	Lobectomy	190	8	5	8	1
4	13	2.8	2.20	Lobectomy	180	49	4	11	1
5	41	3.9	2.27	Lobectomy	130	26	9	10	1
6	47	4.8	1.73	Lobectomy	150	18	3	9	1
7	88	5.6	1.27	Lobectomy	90	3	4	7	1
8	80	7.5	0.68	Lobectomy	135	3	4	6	2
9	44	5.4	2.40	Lobectomy	40	5	2	5	2
10	89	3.5	4.12	Atypical resection	145	4	4	12	1
11	80	6	1.18	Segmentectomy	120	4	3	8	1
12	21	3.4	3.40	Lobectomy	75	24	3	10	1

Pt, patient; d, days; kg, kilogram; CVR, CPAM volume-ratio; CPAM, congenital pulmonary airway malformation; OPT, operation time; MVT, mechanical ventilation time; ITOTDT, indwelling time of thoracic drainage tube; HST, hospital stay time.

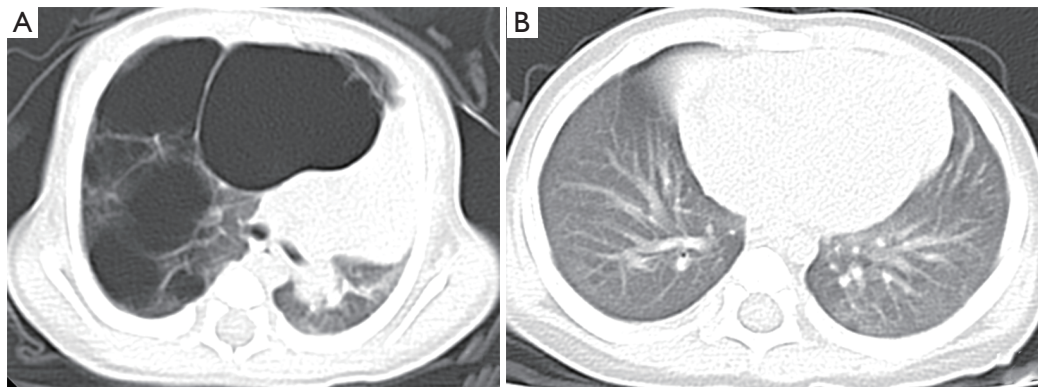


Figure 1 A 13-day-old boy with CPAM in the upper lobe of the right lung, Stocker type 1. CT plain scan images showed that: (A) the giant vesicle caused the heart to move to the left; (B) 1 year after resection of the upper lobe of the right lung, there was no residual lesion, and the heart returned to its original position. CPAM, congenital pulmonary airway malformation; CT, computerized tomography.

hole method was adopted. The 5th intercostal chest in the anterior axillary line was selected as the observation well. The 8th intercostal chest in the midline axillary line and the 5th intercostal chest in the posterior axillary line were usually selected as the operation holes. The operation holes were appropriately adjusted according to the lesion position.

We chose two 5-mm Trocars and one 10-mm Trocar. In children with lobectomy or segmentectomy, LigaSure (Ethicon Endo-surgery, LLC, Somerville, NJ, USA) was used to free the pulmonary arteries, veins, and bronchi,

clamped with absorbable clips, and cut off with an ultrasonic scalpel (Figures 5-7). Notably, the proximal bronchial margin was ligated with non-absorbable silk thread. The boundary between normal and diseased lung tissue in children with atypical resection was clear. Firstly, the edge of diseased lung tissue was marked with an electric hook, and the diseased tissue was removed along the boundary using an ultrasonic scalpel or Endo-GIA (Covidien LLC, Mansfield, MA, USA). Children with repeated pneumonia had severe adhesions in the chest cavity. Thus, it was necessary to use an electric hook

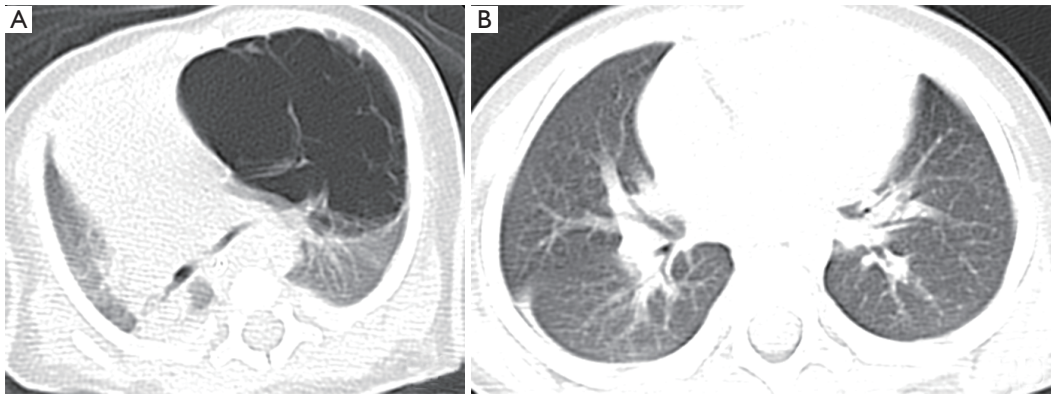


Figure 2 A 47-day-old boy with CPAM in the upper lobe of the left lung, Stocker type 1. CT plain scan images showed that: (A) the giant vesicle caused the heart to move to the right; (B) 1 year after resection of the upper lobe of the left lung, there was no residual lesion, and the heart returned to its original position. CPAM, congenital pulmonary airway malformation; CT, computerized tomography.

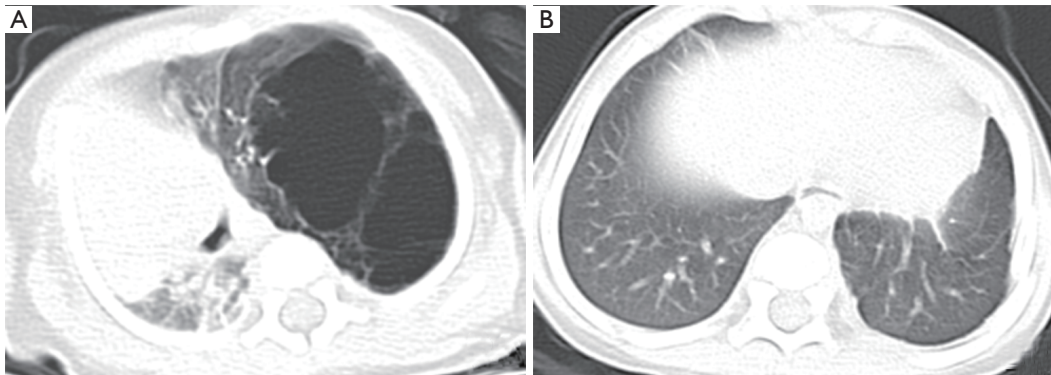


Figure 3 A 31-day-old boy with CPAM in the upper lobe of the left lung, Stocker type 1. CT plain scan images showed that: (A) the giant vesicle caused the heart to move to the right; (B) 1 year after resection of the upper lobe of the left lung, there was no residual lesion, and the heart returned to its original position. CPAM, congenital pulmonary airway malformation; CT, computerized tomography.

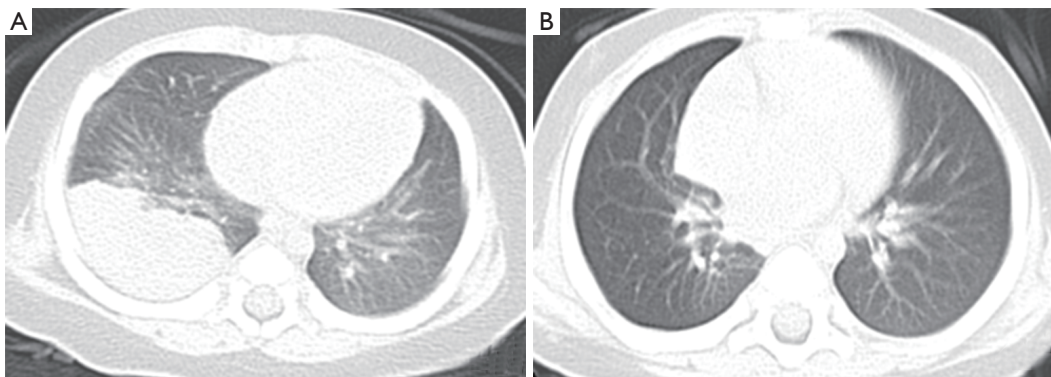


Figure 4 A 85-day-old boy with CPAM in the lower lobe of the right lung, Stocker type 1. CT plain scan images showed that: (A) repeated infection in the lower lobe of the right lung led to lung consolidation; (B) 1 year after resection of the lower lobe of the right lung, there was no residual lesion and inflammation disappeared. CPAM, congenital pulmonary airway malformation; CT, computerized tomography.

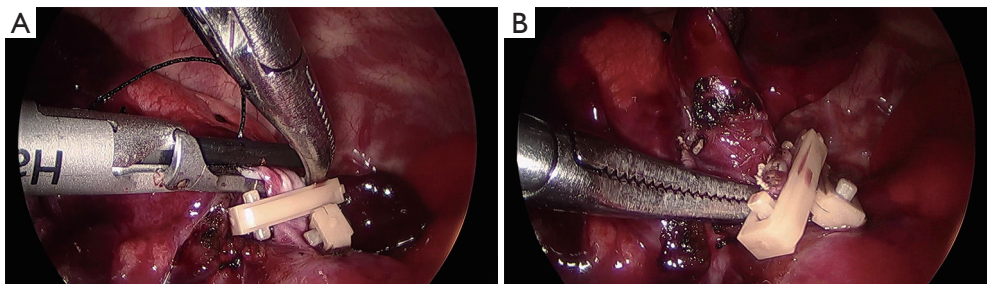


Figure 5 A 21-day-old boy with congenital pulmonary airway malformation in the lower lobe of the right lung. Images taken during thoracoscopic surgery showed that: (A) ligating the artery which supplies the lower lobe of the right lung with an absorbable clip; (B) disconnecting the artery with ultrasonic scalpel.

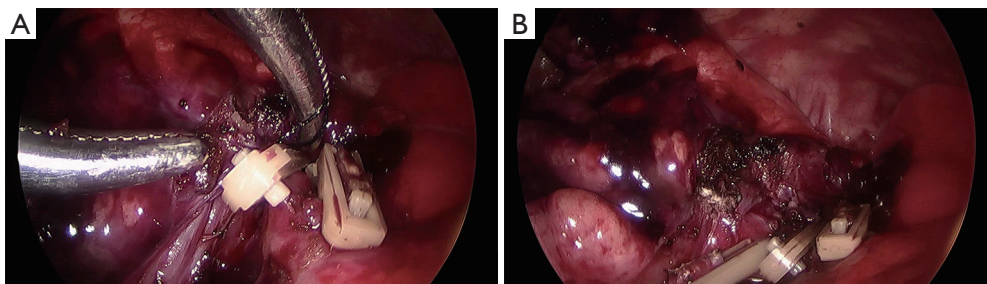


Figure 6 A 21-day-old boy with congenital pulmonary airway malformation in the lower lobe of the right lung. Images taken during thoracoscopic surgery showed that: (A) Ligating the trachea in the lower lobe of the right lung with an absorbable clip; (B) disconnecting the trachea with ultrasonic scalpel.

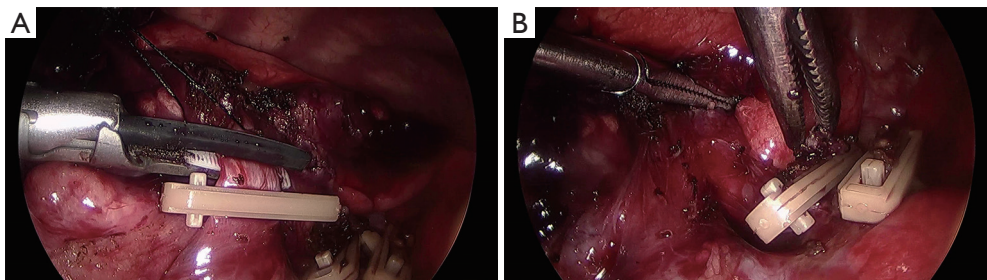


Figure 7 A 21-day-old boy with congenital pulmonary airway malformation in the lower lobe of the right lung. Images taken during thoracoscopic surgery showed that: (A) Ligating the vein returning to the lower lobe of the right lung with an absorbable clip; (B) disconnecting the vein with ultrasonic scalpel.

to free the adhesion between lung tissue and chest wall and expose the affected lung before lobectomy, segmentectomy, or lung-sparing resection.

Statistical analysis

The SPSS22.0 software was used to organize and analyze the data. Results are presented as mean and standard deviation (SD).

Ethical statement

The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This study was approved by the Ethical Committee of the Shanghai Children's Hospital (No. 2023R064-E01). The informed consent was waived by the Medical Ethics Committee of the Shanghai Children's Hospital due to the retrospective design of the study.

Results

All the children underwent VATS, and no cases were converted to thoracotomy. All cases were completed by the same surgeon and the same chief anesthesiologist. The operation time ranged from 40 to 190 minutes, with an average of 124.17 ± 42.74 minutes. In this study, ten children underwent lobectomy, one underwent segmentectomy whose lesion was located in the posterior segment of the upper lobe of the left lung and the scope of the lesion was limited, and one underwent lung-sparing resection whose lesion was located both in the left upper lung and in the left lower lung. Among them, the scope of resection included two cases of left upper lobe, three cases of left lower lobe, two cases of right upper lobe, two cases of right lower lobe, one case of right upper lobe and middle lobe, one case of posterior segment of left upper lobe, and one case of irregular resection of left upper lobe and left lower lobe.

Postoperative mechanical ventilation time ranged from 3 to 49 hours, with an average of 12.75 ± 14.16 hours. Ventilatory weaning was performed when the children were awake from anesthesia, spontaneous breathing resumed, and arterial blood gas analysis showed that there was no carbon dioxide retention (arterial carbon dioxide partial pressure lower than 50 mmHg). The indwelling time of the thoracic drainage tube ranged from 2 to 9 days, averaging 3.92 ± 1.78 days. The postoperative hospital length of stay ranged from 5 to 12 days, averaging 8.42 ± 2.15 days. All these children were hospitalized for a few more days because of their young age, and they were discharged after ensuring that there were no signs of discomfort. Furthermore, postoperative pathological examination showed that nine cases were type 1, two were type 2, and one was type 3.

There were no postoperative complications such as bleeding, bronchopleural fistula, or atelectasis. Postoperative pulmonary air leakage occurred in one case. The child's lesion involved the right upper lobe, and the boundary between the right and middle lobe was unclear. The right upper lobe was resected during the operation, and the wound was large. Moreover, the lung air leakage disappeared after adjusting the drainage tube's position and prolonging the thoracic drainage tube's indwelling time (the total time of indwelling the tube was 9 days).

The follow-up time ranged from 3 months to 4 years. All the children recovered well, with no serious respiratory infection or pneumonia except occasional slight upper respiratory infection during the follow-up. And all of them received chest CTs for about 1 year after operation,

results showing that no residual lesions were found in the reexamined chest CT; the exudation of patchy inflammation in lung disappeared; neither lung consolidation nor lung atelectasis was found; there was no apparent residual cavity in the affected chest. In addition, electrocardiogram (ECG), echocardiography and pulmonary ventilation function test or other examinations were performed if necessary.

Discussion

In 1949, CPAM was called congenital cystic adenomatoid malformation (CCAM), the lung's most common congenital cystic malformation. In 2002, Stocker (1) renamed the disease as CPAM according to its origin, pathological features, and clinical features and divided it into five types according to histological manifestations: 0, 1, 2, 3 and 4.

Children with asymptomatic CPAM after birth do not need immediate surgery. Notably, Kulaylat reports that children with CPAM operated before 3 months of age have a higher incidence of complications than those operated after 3 months of age (3). An imaging examination should be performed during the follow-up after birth for those with prenatally identified lesions by ultrasound, and it is necessary to comprehensively consider the radiation dose for the child and the family's assessment of the risk and choose the operation according to the change of the condition. Verhalleman indicated that in his university hospital, if possible, the first 3 months of life are preserved for lung growth and general development (4). Elective surgery in patients with a high risk for clinical symptoms is recommended before 12 months of age, although no specific guidelines exist (5-8). Referring to the opinions of many scholars, it is suggested that the operation should be performed between 3 months and 1 year old (9-13). So when the children have persistent tachypnea or recurrent pneumonia, early surgery can be considered. Additionally, a few children were born with serious respiratory and circulatory disorders due to the compression of the heart and lungs and mediastinal deviation. Most of these children have huge cystic lesions and emergency surgery should be carried out in time after birth. In our study, all the children had severe respiratory symptoms. Some of them had persistent tachypnea owing to the compression of the heart and lungs; some had recurrent pneumonia; some even required mechanical ventilation because of respiratory failure. So all the surgeries were emergent.

For children under 3 months old with CPAM, thoracotomy is often selected, and good short-term

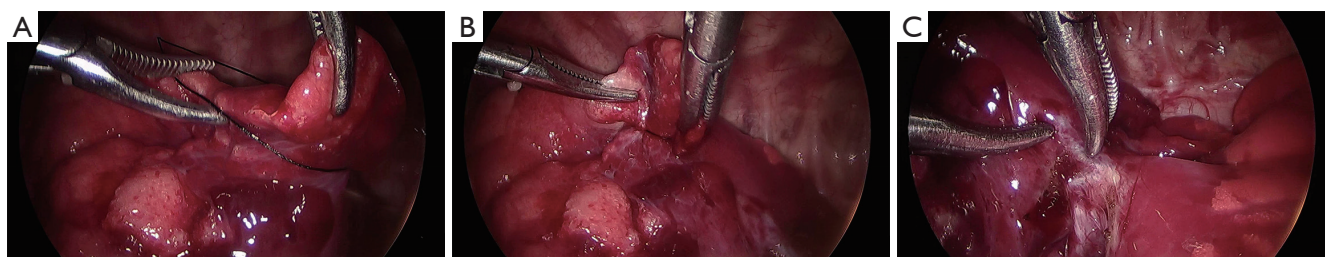


Figure 8 Suspending diseased lung tissue and exposing hilum during thoracoscopic surgery.

results have been observed. However, it also greatly increases the risk of thoracic and/or spinal deformity (14,15). Because the skeletal muscle of early infants is in the development stage, muscle atrophy may occur after cutting the latissimus dorsi and/or serratus anterior muscle in posterolateral thoracotomy, leading to deformity (16). With the improvement of minimally invasive technology and anesthesia management, it is possible to treat CPAM in infants under 3 months old by thoracoscopy. Compared with thoracotomy, VATS has the advantages of smaller incisions, less drainage time, and shorter hospitalization time (17-19). However, there are few related studies at home and abroad because thoracoscopic surgery for infants under 3 months old has narrow chest space. Excessive stretching and squeezing of lung tissue will cause respiratory and circulatory instability. Therefore, how to improve the narrow operating space as much as possible to ensure the stability of the vital signs of the children is the key to the operation's success.

In this study, all the children did not switch to thoracotomy, and the surgical effect was satisfactory. We have some experience in improving the surgical operation space. First and foremost, one-lung ventilation is necessary to obtain sufficient visualization and anatomical space. At present, the bronchial occluder is mainly used to selectively occlude the main bronchus of the affected side to achieve one-lung ventilation. Anesthesiology collaboration is required because the patient's trachea is small under 3 months of age, and it is difficult to place the occluder. Moreover, the left and right main bronchi are short, and the placed occlude easily migrates into different aspects of the airway. If the occlude is dragged into the main bronchi to cause blockage, it will cause acute hypoxia, which will be life-threatening in severe cases. Due to the accumulated clinical experience of bronchial occlusion in children with CPAM, all 12 children in this group were successfully occluded. Usually, the anesthesiologist will first keep the

children in the supine position and place the bronchial occluder under endoscope, then make them in the lateral position. It is important to observe the bronchial occluder under endoscope immediately after changing the body position to ensure that there is no migrations. Even if it is displaced, it is relatively easy to adjust. It is difficult to place bronchial occluder in the lateral position. Additionally, the anesthesiologist closely monitored the respiratory condition during the operation, and there was no case that the occluder was removed due to respiratory instability. Second, artificial pneumothorax can gain more operating space, but too high pressure will affect the vital signs of children. For children less than 3 months of age, we suggest setting the pressure to below 5 mmHg. Third, for artificial pneumothorax and suspension of lung tissue, in the view of the thoracoscope, select a suitable position in the intercostal space on the skin, pull out the needle after the needle silk thread is punctured into the chest cavity, and suspend the diseased lung tissue on the chest wall with the silk thread to expose the hilum, which is convenient for operation (Figure 8). Fourth, if possible, use the electric hook to cauterize the huge vesicles which look like balloons under the enlarged visual field of thoracoscope, so as to reduce the volume of the lesion and increase the operating space (Figure 9). However, attention should be taken not to touch the blood vessels to avoid bleeding affecting the surgical visual field.

Fetal edema is the leading risk factor for poor prognosis of children with CPAM, and the occurrence of fetal edema is directly related to the size of the CPAM. Thus, the risk of fetal edema is often predicted by evaluating the size of the focus in clinical practice. CVR is the specific measurement index, and the calculation formula is (length \times height \times width \times 0.523 of the lesion)/head circumference in centimeter. The larger the CVR value, the larger the relative volume of the lesion and the more pronounced the thoracic space-occupying effect. Notably, when CVR

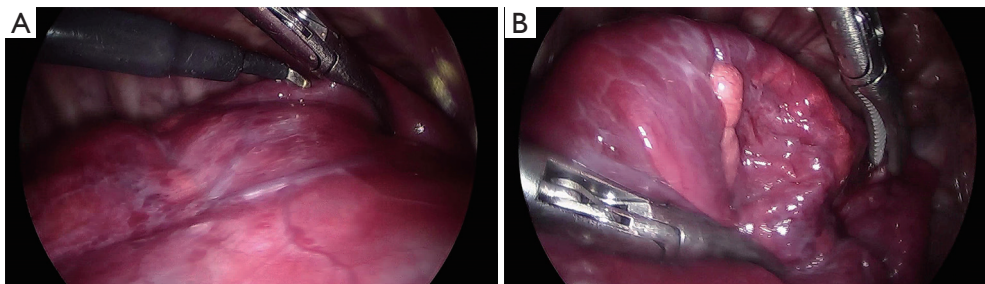


Figure 9 Cauterizing the vesicle with an electric hook to increase the operating space during thoracoscopic surgery.

>1.6, the risk of edema in the fetus of children with CPAM increases, Timothy reports those with CVRs greater than 1.6 have the greatest risk of edema, death, and possible fetal intervention (20). Ocal and his colleagues' research also predicts early surgery with high sensitivity and specificity when getting a threshold value of 0.76 for CVR (21). In this study, the children with CVR >1.6 did not have obvious edema during pregnancy, so no cases were undergoing intrauterine intervention, such as thoraco-amniotic shunt placement. Furthermore, this study shows that although the prognosis of fetal edema is not good, the incidence of fetal edema is low. However, there are few statistical studies on the incidence of fetal edema in China, and a large sample and multi-center study are needed. In addition, we hold that CVR value is not the primary factor to determine the timing of operation, but it is an important factor.

The limitation of this study was that the number of cases in this study was small, and it could only provide limited treatment experience. It is necessary to accumulate a large number of cases to improve the treatment technology further. Additionally, the follow-up time of this study was short. Although the effect is good during the follow-up period, the long-term effect outside the follow-up period is unknown. In the future follow-up process, we will pay close attention to the children's lung development and lung function.

Conclusions

In short, infants with CPAM accompanied by severe clinical symptoms less than 3 months of age as discussed in the preceding content should be operated as soon as possible. VATS is an alternative surgical approach. With the improvement of minimally invasive technology and anesthesia management, VATS is relatively safe and effective in treating children with such conditions.

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Footnote

Reporting Checklist: The authors have completed the STROBE reporting checklist. Available at <https://tp.amegroups.com/article/view/10.21037/tp-23-475/rc>

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://tp.amegroups.com/article/view/10.21037/tp-23-475/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). This study was approved by the Ethical Committee of the Shanghai Children's Hospital (No. 2023R064-E01). The informed consent was waived by the Medical Ethics Committee of the Shanghai Children's Hospital due to the retrospective design of the study.

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