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Impact of pulmonary infection on thoracoscopic surgery outcomes in children with CPAM: a retrospective study

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Abstract

Background Pulmonary infection is a common clinical complication in children with congenital pulmonary airway malformation (CPAM). Surgical intervention has been proven effective in managing this condition. We aim to evaluate the impact of pulmonary infection on the outcomes of total thoracoscopic procedures in children with CPAM.

Methods This was a single-center retrospective study. CPAM patients who underwent total thoracoscopic surgery at a tertiary care center from January 2013 to December 2023 were divided into three groups based on pulmonary infection status: non-infection (NI), hidden infection (HI), and pulmonary infection (PI). Clinical characteristics and operation-related outcomes were compared among the groups.

Results A total of 154 children with CPAM who underwent thoracoscopic surgery were categorized into three groups based on pulmonary infection: the NI group (27 cases), HI group (56 cases), and PI group (71 cases). The conversion rate to thoracotomy was 14.8%, 23.2%, and 29.2% respectively across the three groups. 116 cases were successfully completed thoracoscopically. There were no significant differences in gender distribution among the three groups ($p > 0.05$), but statistically significant ($p < 0.05$) in age and weight. Significant differences emerged in operative time and blood loss ($p < 0.05$), but no significant variations were found in transfusion requirements, chest tube duration, ventilator use duration, or hospital stay length ($p > 0.05$). In postoperative pulmonary complications, a statistically significant difference was found regarding pneumothorax incidence among all three groups ($p < 0.05$), whereas no significant differences ($p > 0.05$) emerged concerning atelectasis or pneumonia incidences across these cohorts.

Conclusions Pulmonary infection is the most prevalent complication in CPAM and exhibits a high rate of hidden infections, thereby complicating surgical intervention and increasing associated risks. Early thoracoscopic intervention prior to infection manifestation can optimize surgical outcomes and reduce associated complications.

Keywords Congenital pulmonary airway malformation, Total thoracoscopic surgery, Pulmonary infection, Children

Introduction

Congenital pulmonary airway malformation (CPAM), formerly known as congenital cystic adenomatoid malformation (CCAM), is historically regarded as a rare condition, with an estimated prevalence ranging from 1/35,000 to 1/7,200 live births [1, 2]. With advancements in medical technology, the reported prevalencehas has shown an upward trend. Prenatal ultrasonography is the primary modality for identifying these lesions, while postnatal CT scans provide definitive diagnosis [3, 4]. Although

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children with CPAM often remain asymptomatic, pulmonary infection is the most common complication. The evolution of surgical techniques has positioned endoscopic surgery as a mainstream therapeutic option for CPAM. Currently, surgical resection of the lesion is the most effective treatment for CPAM. For symptomatic children, the choice between elective surgery and limited surgery is guided by the severity of their clinical presentation [5, 6]. In asymptomatic cases, a divergence of opinion exists among experts. some experts suggest that CPAM may undergo spontaneous regression, suggesting that early surgical intervention could introduce unnecessary risks and advocating for a watchful waiting strategy. In contrast, a substantial body of research supports surgical resection as a safe and effective approach, emphasizing its role in preventing complications and reducing the potential for malignant transformation [7–9].

Previous studies have primarily focused on comparing open surgery and thoracoscopic approaches, with limited attention given to the impact of pulmonary infections on surgical outcomes. In this study, we conducted a review of CPAM cases treated with total thoracoscopic surgery at a single center. Our analysis summarizes the clinical characteristics of these cases and evaluates the influence of pulmonary infections on surgical procedures. It offers valuable insights to inform clinical decision-making and improve patient management strategies.

Materials and methods

This retrospective study was approved by the Children's Hospital of Chongqing Medical University (Approval No. 2023–575). We reviewed the cases of CPAM treated in our center from January 2013 to December 2023. The inclusion criteria were patients who underwent thoracoscopic surgery with a pathological diagnosis of CPAM. Exclusion criteria were CPAM patients who had incomplete clinical data, were aged over 18 years old, or required a second operation due to residual lesions. 154 patients met the inclusion criteria. Patient selection process was shown in Fig. 1.

All patients underwent a preoperative thoracic computed tomographic scan to confirm the diagnosis. According to the site and the size of the lesion, patients underwent total thoracoscopic lobectomy or segmentectomy. Before surgery, cefazolin was given to all patients preventively. General anesthesia was administered using a combination of intravenous and inhalation agents. The patient was positioned in a lateral decubitus position

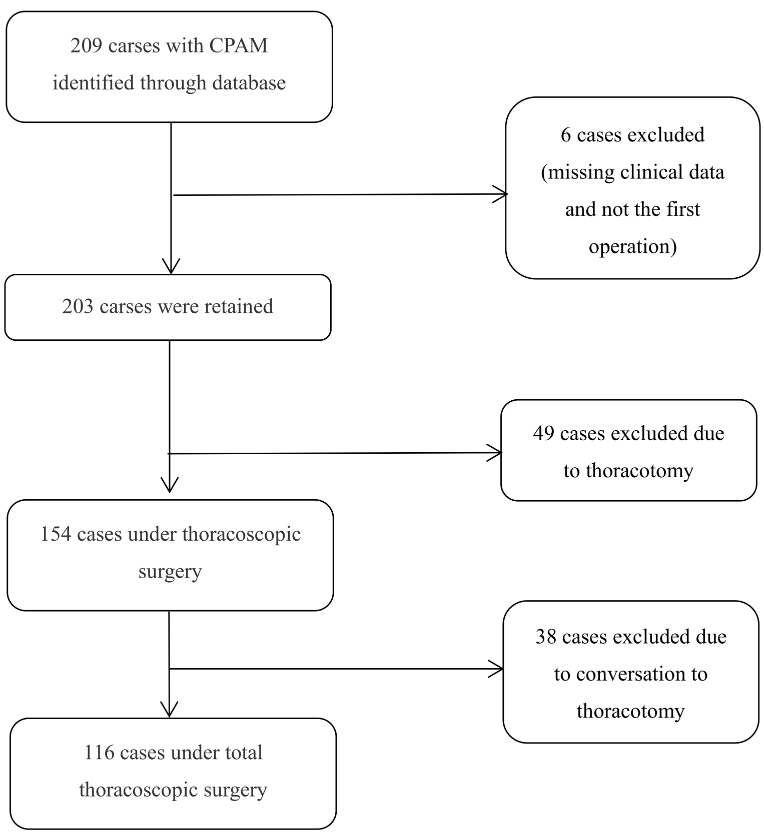

Fig. 1 Flowchart for patient selection

with the unaffected side down. Three thoracoscopic ports were established on the affected side: one at the 6th intercostal space along the anterior axillary line, one at the 7th intercostal space along the posterior axillary line, and one at the 8th intercostal space along the mid-posterior axillary line. An artificial pneumothorax was created to facilitate the procedure. The thoracic cavity was thoroughly explored, and the boundaries of the lesion were marked using an electrocoagulation hook. The corresponding pulmonary ligament was transected, and the associated pulmonary vein was carefully dissected. The pathological lung tissue was resected along the marked boundaries using the electrocoagulation hook, with hemostasis achieved using an ultrasonic scalpel. The relevant bronchus was dissected and clamped to verify normal inflation of the lung lobe. A silicone chest tube was routinely inserted through the 8th intercostal space at the mid-posterior axillary line and connected to a closed thoracic drainage system. After ensuring complete hemostasis, the intercostal incision was closed using a continuous suture with absorbable sutures thread. The muscle and subcutaneous layers were sutured in layers using absorbable sutures. The resected specimen was sent for pathological examination. Following surgery, paraffin sections were prepared from the removed lesion tissue, and the specimens were histologically examined by the pathologist to confirm the diagnosis. To minimize the risk of postoperative complications, postoperative antibiotic therapy was escalated to a third-generation cephalosporin. A thoracic drainage tube was routinely placed during the operation. The drainage tube was removed when the drainage volume was consistently less than 20 mL for three consecutive days.

Children were divided into the NI (Non-infection) group, the HI (hidden-infection) group and the PI (pulmonary infection) group. The classification of the three groups was determined based on postoperative pathological findings. Patients were assigned to NI group if their postoperative pathology showed no evidence of inflammatory cell infiltration (including neutrophils, macrophages, or lymphocytes), regardless of the presence or absence of clinical symptoms. Patients were classified into HI group if their postoperative pathology indicated infection, and they exhibited clinical symptoms such as recurrent fever, cough, wheezing, or elevated inflammatory markers. Additionally, chest radiographs or CT scans showing lesions consistent with infection supported this classification. The remaining patients were categorized into the HI group,as they exhibited no clinical symptoms but with the presence of infection in the postoperative pathology. Consequently, there was a clear distinction between the three groups without overlap. Demographic data of the patients, including age, gender, body weight, lesion side and pathology of the lesion, operative data, and postoperative outcomes were extracted and analyzed statistically.

SPSS 27.0.1 was utilized for statistical analysis in this study. continuous variables were tested for normality. None of the data conformed to a normal distribution, so they were expressed as Md(P25, P75). Differences among three groups were analyzed using the Kruskal–Wallis H test. Categorical data were expressed as percentages (%). Differences among groups were analyzed using the chi-square test. A p-value < 0.05 was considered statistically significant (Fig. 2).

Results

A total of 154 children underwent thoracoscopic surgery in our hospital. They were divided into three groups including the NI group(27 cases), HI group(56 cases), and PI group(71 cases). Among these, 38 cases required conversion to thoracotomy. The rates were 14.8% (4/27) in the NI group, 23.2% (13/56) in the HI group, and 29.2% (21/71) in the PI group. The chi-square test showed no statistically significant differences among three groups ($p = 0.302$). Excluding those that required conversion to thoracotomy, a total of 116 cases were under total thoracoscopy. These included 23 patients in the NI group, 43 in the HI group, and 50 in the PI group. The median age was 21.2 months (IQR, 8.5,66.9 months), and 52.6%

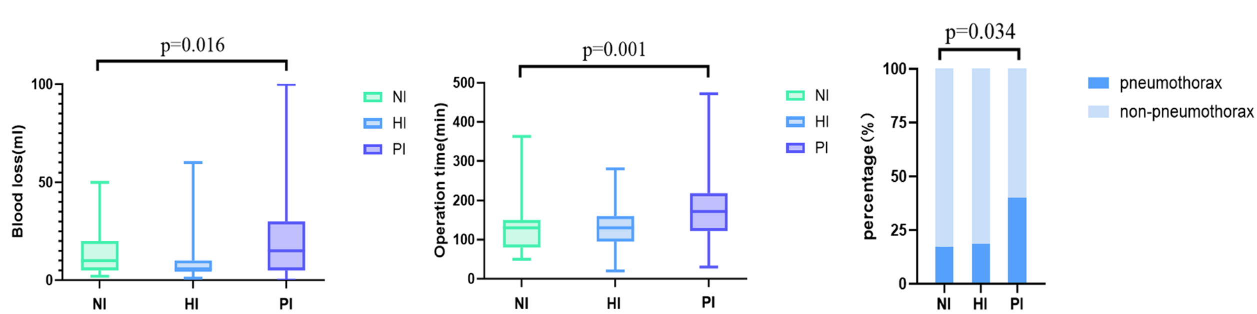

Fig. 2 Distribution of operation time, blood loss and the incidence of pneumothorax among NI,HI and PI groups

were male. The median weight at operation was 11.0 kg (IQR, 8.0,18.0 kg). The differences in sex ($p=0.646$) were not statistically significant but in age ($p<0.001$) and weight ($p=0.004$) were statistically significant among three groups. The majority of lesions were confined to a single lobe. Only 3 cases involved multiple lobes. The left and right lower lobes were the most commonly affected, each with 43 cases (37.1%). Based on the Stocker classification, type 2 CPAM had the highest proportion with 62 (53.4%) cases. Type 1 CPAM had 51 (44.0%) cases. Type 3 and 4 CPAM were rare, with only 2 (1.7%) and 1 (0.9%) cases, respectively. 96 (82.8%) patients underwent thoracoscopic lobectomy and 20 (17.2%) underwent thoracoscopic segmentectomy. The above data was summarized in Table 1.

For the operative findings, PI group had longer operation time, greater blood loss, and more transfusions. Operation time ($p=0.001$) and blood loss ($p=0.016$) showed significant differences among the three groups. To strengthen the robustness of the results, we calculated the effect sizes and confidence intervals by Dunn's post-hoc test for pairwise comparisons. These analyses further support our conclusion. Operation time (NI vs. HI: Cohen's d, 0.12, 95% CI, -20.0 to 20.0 NI vs. PI: Cohen's d, 0.56, 95% CI, 20.0 to 64.0 HI vs. PI: Cohen's d, 0.52, 95% CI, 20.0 to 64.0) and blood loss (NI vs. HI: Cohen's d, 0.32, 95% CI, 0.0 to 10.0 NI vs. PI, Cohen's d, 0.28, 95% CI, 0.0 to 10.0HI vs. PI: Cohen's d, 0.45, 95% CI, 5.0 to 15.0) in the PI group were higher than those in the NI and HI groups, with these differences being statistically significance. No significant differences were found in blood transfusions ($p=0.351$), chest tube duration ($p=0.246$), duration of ventilator use ($p=0.424$), and hospital stay ($p=0.080$). Postoperative complications included atelectasis, pneumothorax, and pneumonia. Atelectasis occurred in 8 cases with 2 (4.7%) in the HI group and 6 (12.0%) in the PI group. Pneumothorax occurred in 32 cases with 4 (17.4%) cases in NI group, 8 (18.6%) cases in HI group, and 20 (40.0%) cases in PI group. Pneumonia occurred in 7 cases with 1 (4.3%) case in NI group, 2 (4.7%) case in HI group, and 4 (8.0%) cases in PI group. There was a significant difference in the incidence of pneumothorax ($p=0.034$) among three groups. However, no significant differences were found for atelectasis ($p=0.064$) or pneumonia ($p=0.740$). For categorical variable, we used odds ratio (OR) as the effect size, and the confidence intervals were calculated using logistic regression. PI group had a significantly higher incidence

Table 1 Patient population and characteristics

Variables	CPAM ($n=116$)	NI ($n=23$)	HI ($n=43$)	PI ($n=50$)	$H/\chi2$	p-value
Age,month,median (IQR)	21.2(8.5,66.9)	15.0(8.6,39.7)	13.1(7.5,25.6)	47.3(15.1,86.0)	$H=13.245$	<0.001[a]
Weight,kg,median (IQR)	11.0(8.0,18.0)	11.0(8.0,16.0)	10.0(7.5,12.5)	15.0(9.5,22.0)	$H=11.008$	0.004[a]
Sex						
Male, *n* (%)	61(52.6)	11(47.8)	25(58.1)	25(50.0)	$\chi2=0.875$	0.646[b]
Female, *n* (%)	55(47.4)	12(52.2)	18(41.9)	25(50.0)		
Lobe						
RUL, *n* (%)	14(12.1)	5(21.7)	3(7.0)	6(12.0)	$\chi2=3.078$	0.215[b]
RML, *n* (%)	7(6.0)	2(8.7)	2(4.7)	3(6.0)	$\chi2=0.432$	0.806[b]
RLL, *n* (%)	43(37.1)	10(43.5)	18(41.9)	15(30.0)	$\chi2=1.899$	0.387[b]
LUL, *n* (%)	6(5.1)	1(4.3)	4(9.3)	2(4.0)	$\chi2=1.290$	0.525[b]
LLL, *n* (%)	43(37.1)	5(21.7)	16(37.2)	22(44.0)	$\chi2=3.347$	0.188[b]
ML, *n* (%)	3(2.6)	None	1(2.3)	2(4.0)	$\chi2=1.019$	0.601[b]
Stocker type						
0, *n* (%)	None	None	None	None		
1, *n* (%)	51(44.0)	21(91.3)	14(32.6)	16(32.0)	$\chi2=26.099$	<0.001[b]
2, *n* (%)	62(53.4)	2(8.7)	28(65.1)	32(64.0)	$\chi2=23.104$	<0.001[b]
3, *n* (%)	2(1.7)	None	1(2.3)	1(2.0)	$\chi2=0.518$	0.772[b]
4, *n* (%)	1(0.9)	None	None	1(2.0)	$\chi2=1.331$	0.514[b]
Total thoracoscopic surgery						
lobectomy, *n* (%)	96(82.8)	19(82.6)	40(93.0)	42(84.0)	$\chi2=2.179$	0.336[b]
segmentectomy, *n* (%)	20(17.2)	4(17.4)	3(7.0)	8(16.0)		

NI Non infection, *HI* Hidden infection, *PI* Pulmonary infection, *RUL* Right upper lobe, *RML* Right middle lobe, *RLL* Right lower lobe, *LUL* Left upper lobe, *LLL* Left lower lobe, *ML* Multiple lobes

[a] Kruskal–Wallis H test

[b] χ^2 test

of pneumothorax (NI vs. HI: OR, 0.92, 95% CI, 0.25 to 3.38 NI vs. PI: OR, 0.32, 95% CI, 0.09 to 1.09 HI vs. PI: OR, 0.34, 95% CI, 0.13 to 0.89) compared to the NI and HI groups. The characteristics of the three groups were shown in Table 2.

Discussion

Lesion resection is a key treatment for children with CPAM, offering both curative effects and reduced complication rates. Although the exact risk remains unquantified, CPAM carries a potential for malignant transformation as evidenced by case reports [10–12]. Surgical intervention may help prevent this potential malignancy. Thoracotomy has a wide range of indications. When patients exhibit respiratory distress, severe thoracic adhesions, or cannot tolerate one-lung ventilation techniques, open surgery ensure optimal outcomes and patient safety. However, thoracoscopy has emerged as the preferred surgical approach due to minimal invasiveness, superior intraoperative visualization, faster recovery, reduced postoperative pain, and shorter hospital stays. In addition, thoracoscopy decreases musculoskeletal impact and effectively lowers complication rates, including scapular winging [13–15].

Segmentectomy is used in clinical as a lung preservation operation to improve postoperative lung function. But its anatomical complexity and variability make it challenging. When the lesion affects multiple lung segments, determining the resection margin becomes difficult. Studies highlight that surgeons must have detailed knowledge of lung segment anatomy to perform segmentectomy successfully [16, 17]. Preoperative CT imaging is used to localize and qualitatively diagnose the disease to guide surgical planning. During surgery, careful exploration of the lesion and anatomical details could reduce the risk of leaving residual tissue behind [17–19]. Lobectomy is the standard procedure for treating CPAM in children. If segmental dissection is difficult or complete lesion removal is uncertain, switching to lobectomy can lower the risk of complications and avoid prolonged surgery [18, 20]. In this study, 116 patients underwent total thoracoscopic surgery including 96 lobectomies and 20 segmentectomies. All segmentectomy patients received preoperative CT 3D reconstruction to identify the specific lung segments and subsegments for removal. The severed bronchus and arteriovenous were determined intraoperatively based on reconstructed images and actual anatomy. With the progress of surgical technology, the use of segmentectomy in our hospital has steadily increased.

CPAM is diagnosed by prenatal ultrasound and postpartum CT before symptoms develop. Nevertheless, the majority of patients opt for surgery after symptoms appear. It was reported that the rate of pulmonary infection increases with age, especially after 2 years old [8]. Our data support this finding, showing that all patients in the PI group exhibited infection symptoms, with older age and higher weight being common characteristics. Regarding lesion location, we found no significant differences among three groups. Most lesions were localized to a single lobe, with the lower lobe being the most frequently affected [16, 18]. Notably, type 2 CPAM accounted for the largest proportion within the PI group, showing a significant difference. According to Stocker classification, type 2 CPAM is located in the bronchi and bronchioles, consists of multiple cysts, and primarily present with pulmonary infection as their main clinical feature [21, 22]. Hermelijn SM et al. found a correlation between lesion size and infection rate, based on pulmonary anatomical characteristics. Inflammation was more common in CPAM lesions with low gas content and small volume [18]. This results were consistent with ours.

Table 2 Comparison of related variables among NI,HI and PI groups

Variables	NI ($n=23$)	HI ($n=43$)	PI ($n=50$)	$H/\chi2$	p-value
Operation time,min,median (IQR)	130.0(80.0,150.0)	130.0(95.0,160.0)	172.0(122.3,218.5)	$H=13.306$	$p^a=0.001$
Blood loss,ml,median (IQR)	10.0(5.0,20.0)	5.0(5.0,10.0)	15.0(5.0,30.0)	$H=8.316$	$p^a=0.016$
Blood transfusions, n (%)	None	2(4.7%)	4(8.0%)	$\chi2=2.093$	$p^a=0.351$
Chest tube duration,day,median (IQR)	4.0(3.0,5.0)	4.0(3.0,5.0)	6.0(2.8,8.3)	$H=2.804$	$p^a=0.246$
Duration of ventilator use,day,median (IQR)	2.0(0.5,5.0)	3.0(1.0,5.0)	3.0(1.0,4.0)	$H=1.715$	$p^a=0.424$
Hospital stay,day,median (IQR)	13.0(11.0,17.0)	15.0(13.0,20.3)	15.0(12.0,17.0)	$H=5.062$	$p^a=0.080$
Postoperative complications					
atelectasis, n (%)	None	2(4.7%)	6(12.0%)	$\chi2=4.069$	$p^b=0.064$
pneumothorax, n (%)	4(17.4%)	8(18.6%)	20(40.0%)	$\chi2=6.790$	$p^b=0.034$
pulmonary infection, n (%)	1(4.3%)	2(4.7%)	4(8.0%)	$\chi2=0.601$	$p^b=0.740$

[a] Kruskal–Wallis H test

[b] χ^2 test

Our data show that a history of pulmonary infection impacts the outcome of thoracoscopic procedure. NI group had shorter operation time, fewer blood transfusions, and reduce blood loss. Specifically, the median surgical time in the NI group was 130 min, markedly lower than the 170 min observed in the PI group and 140 min recorded in the HI group. Our operating time for all three groups was lower than those reported in previous studies on thoracoscopic surgery around 180 min [23–25]. PI group with significant pulmonary inflammation presented with thoracic adhesion. This complicated the surgical procedure. This observation is consistent with existing literature. Preoperative pulmonary infections are associated with higher rates of conversion to open surgery, prolonged operative times, and extended postoperative hospital stays. Comparative data analysis revealed that the NI group had fewer intraoperative blood transfusions, shorter durations of ventilator dependency, and reduced hospital stays compared to the HI and PI groups. Although these differences did not achieve statistical significance, they suggest that the NI group may have been associated with lower surgical complexity. The blood loss could be used to evaluate the effect of surgery. We observed significant differences in blood loss. PI group exhibited the highest median intraoperative blood loss (20 ml) and demonstrated the greatest demand for transfusions (8.0%). One study reported no statistical difference in blood transfusion based on age cohort or sample size [26]. While our investigation focused on infection grouping without age stratification. Therefore, we could not assume that the blood loss was age-related. We focused on lung related complications including atelectasis, pneumothorax, and pneumonia. Although only pneumothorax incidence reached statistical significance, complication rates showed a declining trend from the PI to NI groups. Notably, the PI group required longer chest tube placement due to more extensive surgical injury and increased exudate production. Additionally, it heightened the risk of air leakage. This was consistent with other studies reporting higher postoperative complication rates in infected lesions [27–29].

When deciding whether to choose surgical intervention for CPAM children, it is essential to consider the patient's infection status and surgical risks. For children in the HI or NI group, early surgery may be the preferable option. These patients typically present with lower surgical complexity, faster postoperative recovery, and a reduced risk of complications. Early intervention can prevent the exacerbation of infections and reduce the complexity of future surgeries. HI group remains a certain risk of postoperative complications, although the infection is hidden. For PI group, delaying surgery may be more appropriate. Preoperative anti-infective treatment to control inflammatory responses could minimize intraoperative bleeding and tissue adhesion, thereby reducing surgical difficulty and the risk of postoperative complications. To mitigate the risk of postoperative complications, we opted for a routine indwelling thoracic drainage tube. Closely monitor the chest drainage to detect and manage complications such as pneumothorax in a timely manner. Additionally, respiratory function exercises and physical therapy should be implemented to help reduce the incidence of atelectasis and pulmonary infections.

This study has several limitations. First, our data comes from a single center, limited by treatment concept, which may cause selection bias. Second, we focused on the association with infection and surgery, ignoring the difference in the severity of the disease among groups. That might have affected the surgical data. Third, this is a retrospective study, and long-term prognostic outcomes are needed to better explore the effects of infection.

Conclusion

We performed a retrospective study on pulmonary infection in children with CPAM. Lesion excision under thoracoscopic surgery is safe and effective. CPAM is prone to pulmonary infection with a high hidden-infection rate. This increases surgical difficulty and risk. Therefore, thoracoscopic surgery proves to be safe and effective prior to the symptoms. Surgery is recommended in asymptomatic cases for CPAM under 1 year of age. Additionally, such procedures may be deferred until the children reach 2 years of age, depending on surgical conditions and family preferences.

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Not applicable.

Authors' contributions
Yong-Gang Li designed and guided the study. Cui-xia Yuan performed the analysis of the data. Chun Wu and Zheng-Xia Pan helped with data and reference collection. All authors contributed to manuscript writing and revision, and approved the submitted version.

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Data availability
Data is provided within the manuscript.

Declarations

Ethics approval and consent to participate
This retrospective study is in compliance with the Helsinki Declaration and approved by the Children's Hospital of Chongqing Medical University (Approval No. 2023–575). After review by the Ethics Committee of the Children's Hospital Affiliated to Chongqing Medical University on December 23, 2023, this study utilizing medical records and biological specimens obtained from prior clinical treatments was determined not to pose unnecessary risks to participants. Consequently, the committee agreed to waive the requirement for informed consent, provided that patient privacy is strictly protected.

Consent for publication
Not applicable.

Competing interests
The authors declare no competing interests.

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