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Open versus thoracoscopic approach in the surgical treatment of congenital pulmonary airway malformations: A retrospective cross-sectional analysis and review

Jean Dai [a,*], Jesse York [a], Baylor Schexnayder [b], Frankie Fike [b]

[a] Texas Tech Health Sciences Center (TTUHSC) School of Medicine, 3601 4th St, Lubbock, TX 79430, United States
[b] TTUHSC Department of Surgery, Lubbock, TX, United States

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ABSTRACT

Introduction: Congenital pulmonary airway malformations (CPAMs) are rare developmental lung abnormalities that often require surgical intervention. This study aimed to compare resource utilization and in-hospital outcomes between open thoracotomy and thoracoscopic approaches for CPAM treatment.
Methods: We used the Kids' Inpatient Database (KID), the largest publicly available all-payer pediatric inpatient care registry, to identify patients under 20 years old who were treated with elective surgery for CPAM in 2016 and 2019. Patients were categorized into open thoracotomy or thoracoscopic groups based on International Classification of Disease, Tenth Revision (ICD-10) procedure codes. We analyzed differences in demographic characteristics, complications, total cost of stay, and length of hospital stay between the two groups.
Results: The study included 749 patients (436 thoracoscopic, 313 open). Demographic analysis revealed significant differences in sex distribution ($p = 0.028$) and race ($p < 0.001$) between groups. The mean age was similar (thoracoscopic: 1.2 years, open: 1.7 years; $p = 0.059$). Complications were not significantly different between approaches. The thoracoscopic approach was associated with lower mean total cost of stay ($74,719 vs. $82,146; $p = 0.037$) and shorter mean length of stay (3.1 vs. 4.5 days; $p < 0.001$) compared to the open approach (Table 1).
Conclusion: This study suggests that the thoracoscopic approach for CPAM treatment may offer advantages in terms of reduced hospital costs and shorter length of stay without significantly increasing complication rates. These findings could inform clinical decision-making and resource allocation in pediatric surgical care. Further research is needed to assess long-term outcomes and patient-reported measures between these approaches.
Type of study: retrospective cross-sectional analysis
Level of evidence: 4

Background

Congenital pulmonary airway malformations (CPAM) are rare congenital anomalies of the lung leading to cystic and/or adenomatous pulmonary lesions [1]. Currently, the Stocker classification describes five types of CPAM, listed below [1,2].

- Type 0 is incompatible with life and starts in the trachea or bronchi with histology showing bronchial-type airways with cartilage, smooth muscle, and glands separated by mesenchyme.
- Type 1 is the most frequent type, accounting for about 70 % of all CPAM cases. It originates in the bronchi often as multiloculated cysts located within one lobe and lined with pseudo-stratified columnar epithelium. About half of the cases show cellular hyperplasia. The prognosis is excellent after resection; however, there is a very rare chance of malignant transformation to broncho-alveolar carcinoma.
- Type 2 starts in the bronchiolar regions and is the second most frequent type. It is often associated with other congenital anomalies such as renal agenesis, cardiovascular defects, diaphragmatic hernia, skeletal defects, and esophageal atresia. It usually presents as multiple small cysts without a mass effect. The prognosis is good with no malignant potential.
- Type 3 also originates in the bronchiolar regions but is much less common. It typically involves and expands throughout an entire

* Corresponding author.
E-mail address: jean.dai@ttuhsc.edu (J. Dai).

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lobe, compressing the other lobes, and the lesions are usually solid rather than cystic. The prognosis is favorable due to no malignant potential.

- Type 4 is the rarest type and originates in the acinar structures of the lung. They consist of peripheral thin-walled cysts that are multiloculated and lined by alveolar type 1 or type 2 cells. There is a strong association between these CPAMs and transformation to type 1 pleuropulmonary blastomas.

CPAM diagnosis is often performed during prenatal ultrasound imaging. Earlier diagnosis has improved postnatal management strategies and knowledge on the pathophysiology of CPAM. Symptoms are widely variable, ranging from asymptomatic to life-threatening. Untreated CPAMs can lead to recurrent infections, hypoxia, and transformation to malignancy as described by the Stocker classification above. Surgical lobectomy via thoracoscopic or open approach is considered first line for symptomatic cases [3]. Despite advancements in diagnostic methods and surgical approach, there remains limited available knowledge and research due to the rarity of this condition and vulnerability of the pediatric population.

Currently, treatment modality and timing of surgical intervention for asymptomatic cases remains controversial. Recent literature supports that early elective surgical treatment may benefit patients with asymptomatic CPAMs to prevent future complications. This recommendation assumes that waiting for asymptomatic CPAMs to become symptomatic increases the likelihood and rate of infection over time which eventually makes surgery more challenging. However, there is debate on whether the risks of surgery and complications outweigh the benefit of preemptive intervention. Additionally, there is no consensus on best timing or type of surgical approach intervention for asymptomatic patients. Other studies suggest that there is low risk of symptom development in asymptomatic cases, so surgical intervention alone may cause unnecessary risks to patients who would otherwise remain asymptomatic throughout their lives. The exact risk for symptom development is unknown but has been reported to be anywhere from 3 to 24 % based on retrospective analyses, although these studies generally have limited follow-up [4–8].

Due to the conflicting management strategies of asymptomatic CPAMs and limited available research, further investigation is needed. In this study, we provide an analysis and review of recent data of surgical treatment of CPAMs reported in the largest publicly available all-payer pediatric inpatient care registry. Specifically, we aim to further understand the surgical treatment for this rarely studied congenital anomaly in order to provide a more in-depth look into the trends, comorbidities, and demographics of pediatric patients with CPAM.

Methods and materials

We used the Kids' Inpatient Database (KID), the largest publicly available all-payer pediatric inpatient care registry, to identify patients who were treated with elective surgery for CPAM in 2016 and 2019. Institutional Review Board (IRB) approval was not necessary as the KID is a de-identified, publicly available dataset that ensures patient confidentiality. Patients between 0 and 20 years of age with the International Classification of Disease, Tenth Revision (ICD-10) code Q33.0 (Congenital Cystic Lung) who were treated with surgery during an elective hospital admission were included. Cases were weighted per Healthcare Cost and Utilization Project recommendations using the hospital discharge weights provided in the datasets [9].

Patients were categorized into open thoracotomy or thoracoscopic groups based on ICD-10 Procedure Coding System codes (Appendix A). We analyzed differences in characteristics including age, sex, and race, presence of complication(s), total cost of stay, and length of hospital stay (LOS) between the two approaches. Complications were defined by the ICD-10 codes listed in Appendix A.

We then looked at the most frequent additional ICD-10 diagnostic and procedural codes among all CPAM patients who met inclusion criteria (Table 2). Additionally, we analyzed differences in procedural approach, LOS, and presence of complications between patients under 1 year old and those older than 1 year. This cutoff assumes that symptomatic patients diagnosed prenatally were treated within the first year of life. Lastly, we looked into differences in cost between patients under 1 year old and those older than 1 year.

All data lookup, filtering, and summarization were performed using R (version 4.3). All statistical analyses were conducted using IBM SPSS Statistics, Version 29.0 (IBM Corp.). Two-group comparisons were performed with $\chi 2$ and Welch's *t*-tests for categorical and continuous variables, respectively.

Results

749 patients met inclusion criteria. Of these, 313 underwent open surgery and 436 underwent thoracoscopic surgery for treatment of CPAM. Demographic analysis revealed significant differences in sex distribution ($p = 0.028$) and race ($p < 0.001$) between the two groups with males and white, non-Hispanics being more common (Table 1). The mean age was similar in both groups (thoracoscopic: 1.2 years, open: 1.7 years; $p = 0.059$).

Table 1
Analysis of thoracoscopic vs open procedures in CPAM patients.

Characteristic	Thoracoscopic (N = 436)	Open (N = 313)	Study population (N = 749)	*P* value[1]
Age – years				
Mean	1.2	1.7	1.4	
Median	0	0	0	0.059
Range	0–17	0–20	0–20	
Sex – no. (%)				**0.028**
Female	188 (43.1)	160 (51.1)	348 (46.5)	
Male	248 (56.9)	153 (48.9)	401 (53.5)	
Race — no. (%)				**<0.001**
White, non-Hispanic	224 (51.4)	151 (48.2)	375 (50.1)	
African American	49 (11.2)	17 (5.4)	66 (8.8)	
Hispanic	76 (17.4)	70 (22.4)	146 (19.5)	
Asian or Pacific Islander	23 (5.3)	8 (2.6)	31 (4.1)	
Native American	3 (0.7)	0 (0)	3 (0.4)	
Other/Not Identified	61 (14.0)	67 (21.4)	128 (17.1)	
Complications — no. (%)				
Total	75 (17.2)	63 (20.1)	137 (18.3)	0.376
Any	63 (14.4)	58 (18.5)	121 (16.2)	0.132
Procedural complications, unspecified	17 (3.9)	10 (3.2)	26 (3.5)	0.604
Intraoperative and post-procedural respiratory complications	58 (13.3)	53 (16.9)	111 (14.8)	0.168
Total cost of stay (USD)				
Mean	$74,719	$82,146	$77,848	**0.037**
Median	$63,633	$71,522	$66,744	
Minimum	$15,246	$24,554	$15,246	
Maximum	$425,654	$330,224	$424,654	
Length of stay (days)				
Mean	3.1	4.5	3.7	
Median	2.0	3.0	3.0	**<0.001**
Range	28	31	32	

[1] : *P* value was calculated using Pearson's Chi-square test for categorical variables and independent samples *t*-test for continuous variables. **Bolded values** are statistically significant ($P < 0.05$).

Total number of complications were not significantly different between the two groups. However, the thoracoscopic approach was associated with lower mean total cost of stay ($74,719 vs. $82,146; $p = 0.037$) and shorter mean length of stay (3.1 vs. 4.5 days; $p < 0.001$) compared to the open approach. The proportion of cases performed thoracoscopically significantly increased from 2016 to 2019 (from 53.8 % to 62.2 %, $p = 0.0154$). When stratified based on age, there was a significant difference in LOS between infants and older patients (5.1 days vs. 3.1 days, $p < 0.001$), although there were no significant differences in surgical approach or presence of complications between the two groups.

Frequencies of coexisting ICD-10 diagnoses are listed in Table 2, with postprocedural pneumothorax, acute post procedural pain, and pneumothorax being among the 3 most frequent coexisting diagnoses. Frequencies of ICD-10 Procedure Coding System codes are listed in Table 3, where PCS codes including the word "resection" involved wedge resection of a particular lobe, while those including the word "excision" involved lobectomy, or removal of the entire lobe. Results from Table 3 are further depicted in Fig. 1 in order to visualize the differences in rate of thoracoscopic vs. open approach among lung procedures categorized by lobe and laterality.

Discussion

We aimed to compare the characteristics and outcomes of thoracoscopic and open thoracotomy approaches for elective CPAM treatment in pediatric patients using the 2016 and 2019 Kids' Inpatient Database. We found that patients who underwent thoracoscopic surgery had a lower mean total cost of stay ($74,719 vs. $82,146; $p = 0.037$) and a shorter mean LOS (3.1 vs. 4.5 days; $p < 0.001$) compared to those who underwent open surgery. These findings are significant as they suggest that minimally invasive surgery can lead to more efficient use of healthcare resources and quicker recovery times for patients. However, since we have no insight into why a specific approach was chosen for each patient (e.g., surgeon preference, size and characteristics of the lesion, size of the patient, comorbid conditions), we cannot infer that thoracoscopic surgery is superior to open surgery. The lack of a significant difference in complication rates between the two approaches supports the safety of thoracoscopic surgery in this patient population.

Although there are conflicting viewpoints regarding timing of surgical intervention for CPAM, recent literature suggests that earlier resection may be favorable to avoid complications/difficult surgery in the future. Interestingly, we observed a significant difference in LOS between patients under 1 year old and those older, with younger patients having approximately 2 days longer LOS ($p < 0.001$). However, there was no significant difference in surgical approach or complication rates between the two age groups. This longer LOS in younger patients might be attributed to the complexities associated with treating infants, who may have additional medical concerns or require more intensive postoperative monitoring. Although our results might indicate no harm in waiting longer to operate, we would need a more focused study to make any conclusions regarding optimal timing of surgery.

Our data revealed an increasing trend in the use of thoracoscopic surgery from 2016 to 2019 (from 53.8 % to 62.2 %, $p = 0.0154$). This trend likely reflects growing confidence in and familiarity with minimally invasive techniques among pediatric surgeons. It may also indicate a broader acceptance of thoracoscopic approaches as the standard of care for CPAM treatment. However, review of additional ICD-10-PCS codes revealed that there may be differences in operative strategy depending on the area of the lung requiring resection or excision (Fig. 1). Interestingly, rates of percutaneous vs open procedure varied between the area of operation, with resections of the lingula and left lower lobe being more commonly performed thoracoscopically than other resections. Although specific reasoning for these differences cannot be deduced from the data, these findings may indicate a need for further analysis of the effects of anatomical location on the difficulty and

Table 2
Most common additional ICD-10 diagnoses in CPAM patients.

ICD-10 Code	Description	Frequency
Q330	Congenital cystic lung	749
Z8701	Personal history of pneumonia (recurrent)	25
J95811	Postprocedural pneumothorax	24
J939	Pneumothorax, unspecified	22
Z5332	Thoracoscopic surgical procedure converted to open procedure	22
J9811	Atelectasis	21
K219	Gastro-esophageal reflux disease without esophagitis	21
Q332	Sequestration of lung	21
J95812	Postprocedural air leak	20
Q339	Congenital malformation of lung, unspecified	19
J9382	Other air leak	18
Q348	Other specified congenital malformations of respiratory system	18
Z7722	Contact with and (suspected) exposure to environmental tobacco smoke (acute) (chronic)	17
K5900	Constipation, unspecified	16
R000	Tachycardia, unspecified	16
J948	Other specified pleural conditions	15
Q211	Atrial septal defect	15
R0902	Hypoxemia	15

Table 3
30 most common CPAM-related procedures.

PCS Code	Description	Frequency
0BTF4ZZ	Resection of Right Lower Lung Lobe, Percutaneous Endoscopic Approach	113
0BTJ4ZZ	Resection of Left Lower Lung Lobe, Percutaneous Endoscopic Approach	100
0BTF0ZZ	Resection of Right Lower Lung Lobe, Open Approach	84
0BTJ0ZZ	Resection of Left Lower Lung Lobe, Open Approach	55
0BBJ4ZZ	Excision of Left Lower Lung Lobe, Percutaneous Endoscopic Approach	49
0BTG4ZZ	Resection of Left Upper Lung Lobe, Percutaneous Endoscopic Approach	41
0BBF4ZZ	Excision of Right Lower Lung Lobe, Percutaneous Endoscopic Approach	34
0BBH4ZZ	Excision of Lung Lingula, Percutaneous Endoscopic Approach	30
0BBJ0ZZ	Excision of Left Lower Lung Lobe, Open Approach	30
0BTC4ZZ	Resection of Right Upper Lung Lobe, Percutaneous Endoscopic Approach	29
0BTC0ZZ	Resection of Right Upper Lung Lobe, Open Approach	29
0BBF0ZZ	Excision of Right Lower Lung Lobe, Open Approach	28
0BTG0ZZ	Resection of Left Upper Lung Lobe, Open Approach	25
0BTD4ZZ	Resection of Right Middle Lung Lobe, Percutaneous Endoscopic Approach	19
0BTD0ZZ	Resection of Right Middle Lung Lobe, Open Approach	16
0BBG0ZZ	Excision of Left Upper Lung Lobe, Open Approach	14
0BBC4ZZ	Excision of Right Upper Lung Lobe, Percutaneous Endoscopic Approach	8
0BBC0ZZ	Excision of Right Upper Lung Lobe, Open Approach	8
0BBG4ZZ	Excision of Left Upper Lung Lobe, Percutaneous Endoscopic Approach	7
0BBK0ZZ	Excision of Right Lung, Open Approach	4
0BBD4ZZ	Excision of Right Middle Lung Lobe, Percutaneous Endoscopic Approach	3
0BBK4ZZ	Excision of Right Lung, Percutaneous Endoscopic Approach	3
0BBL4ZZ	Excision of Left Lung, Percutaneous Endoscopic Approach	3
0BBD0ZZ	Excision of Right Middle Lung Lobe, Open Approach	4
0BQJ4ZZ	Repair Left Lower Lung Lobe, Percutaneous Endoscopic Approach	3
0BBL0ZZ	Excision of Left Lung, Open Approach	1
0BTH0ZZ	Resection of Lung Lingula, Open Approach	1
0BTL0ZZ	Resection of Left Lung, Open Approach	1
0BU407Z	Supplement Right Upper Lobe Bronchus with Autologous Tissue Substitute, Open Approach	1
0BQC0ZZ	Repair Right Upper Lung Lobe, Open Approach	2

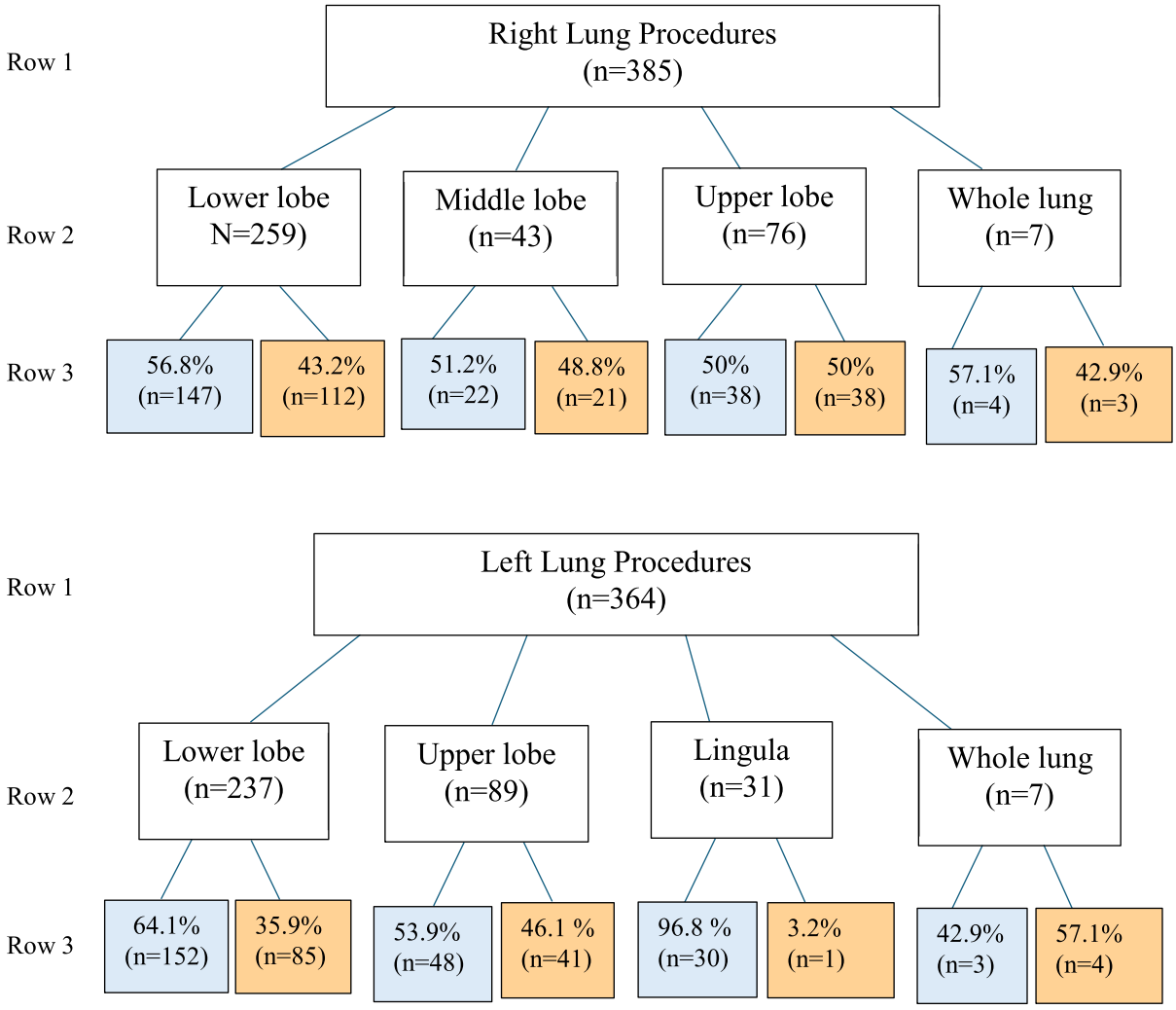

Figure Legend:
Row 1: Number of procedures based on laterality, including wedge resections, lobectomies or excisions, and repairs
Row 2: Lobe of lung being operated on
Row 3: Thoracoscopic approach | Open approach
Percentages (%) in Row 3 are based off of number of procedures for corresponding lung section in Row 2
Row 4:

n=number of procedures
*Figure created by Jean Dai

Fig. 1. Procedure breakdown by lobe and laterality*.

type of procedure performed.

Additionally, our analysis of the most frequent additional diagnostic and procedure codes provide insights into common coexisting conditions and procedures among CPAM patients. Postprocedural pneumothorax, acute postprocedural pain, and unspecified pneumothorax were the top three coexisting conditions, highlighting the respiratory complications often encountered in these patients. However, grouping each complication into broader categories such as "respiratory complications" restricts the ability to assess specific risks and the severity of outcomes. Providing a more detailed breakdown of complications may improve the study's ability to identify more specific risks and outcomes in patients with CPAM.

Furthermore, patient selection bias may not be adequately accounted for in this study. Due to limitations in study design and available information in the KID Database, several factors that were not accounted for in the selection criteria, including the severity of CPAM cases, patient symptoms, and surgeon experience. Additionally, the study did not examine operative time or blood loss, which are critical indicators of surgical burden and complication risk. Including such details in future analyses could provide a more comprehensive understanding of their impact.

Another limitation of the study is the lack of follow-up data, which does not assessment of long-term outcomes, such as the impact of early surgical intervention on postoperative quality of life, reoperation rates,

or other long-term health metrics. These data are vital to validate the benefits of different surgical approaches and to inform optimal timing for intervention.

Lastly, the retrospective design of this study inherently limits control over confounding variables, and as a result, establishing causative relationships between surgical approaches and outcomes is not possible. Future prospective studies that address these gaps by analyzing patient selection criteria, operative details, and long-term outcomes will provide valuable insights to optimize care for patients with CPAM and guide clinical decision-making.

Conclusion

Our study supports existing literature that the thoracoscopic approach for CPAM treatment offers significant benefits in terms of reduced hospital stay and costs without increasing complication rates. These findings underscore the value of minimally invasive surgery in pediatric patients. The trends and outcomes observed in this study could help shape future clinical guidelines and improve the standard of care for pediatric patients with CPAM.

Long-term follow-up studies in asymptomatic CPAM patients are lacking, preventing accurate estimation of long-term symptom development rate in this patient group. Data does not differentiate patients based on whether or not they are symptomatic or asymptomatic.

While our study provides valuable insights, it has several limitations. The use of a retrospective database limits the ability to control for all potential confounding variables, and the reliance on ICD-10 codes may not capture all nuances of patient conditions and surgical details. Future prospective studies could provide more detailed information on patient selection criteria, intraoperative techniques, and long-term outcomes. Additionally, investigating the causes underlying demographic differences in surgical approach could help address potential disparities in care.

CRediT authorship contribution statement

Jean Dai: Writing – review & editing, Writing – original draft, Methodology, Investigation, Data curation. **Jesse York:** Software, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Baylor Schexnayder:** Writing – review & editing, Supervision. **Frankie Fike:** Writing – review & editing, Validation, Supervision, Resources.

Declaration of competing interest

We certify that there is no financial/personal interest or belief that could affect our objectivity. No competing interests exist.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.yjpso.2025.100193.

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