Original Article

Thoracoscopic surgery for rare congenital pulmonary airway malformations in adults: a decade of retrospective study

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Background: Congenital pulmonary airway malformation (CPAM) is a rare benign deformity of the lungs in adults. Our study aimed to evaluate the clinical features and compare the effect of thoracoscopic lobectomy and wedge resection for adult CPAMs.

Methods: This was a retrospective study including eighteen adults with CPAMs recruited between 2013 and 2023. Radiological scans and pulmonary function test (PFT) were performed before operation. All the patients were treated with thoracoscopic approach, which were categorized into groups of lobectomy and wedge resection. The baseline, preoperative, and operative data were evaluated and analyzed.

Results: Four males and fourteen females were diagnosed with CPAMs at a median age of 57.5 years. Cough was the main symptom, reported by 55.6% of the patients. CPAMs were always initially misdiagnosed as other conditions due to heterogeneous computed tomography (CT) characteristics. The mean of PFT results showed normal (>80% predicted) in forced vital capacity (FVC), forced expiratory volume in 1 s (FEV1), and FEV1 to FVC ratio (FEV1/FVC), but less than 70% predicted in forced expiratory flow (FEF) at 25–75%, 50% and 75% of FVC. All patients underwent video-assisted thoracic surgery (VATS) with a total of nine wedge resections and nine lobectomies. Age at surgery varied statistically between the two groups. Whereas, duration of surgery, blood loss, postoperative drainage, days of drainage, days of hospitalization and postoperative complications showed no statistical difference between the two groups. There were 27.8% of the lesions showing CPAMs mixed with other diseases during histological evaluations.

Conclusions: CPAM in adults showed a complex presentation in terms of clinical symptoms, imaging performance and pathological findings. Half of the patients were detected with small airway dysfunction preoperatively. Thoracoscopic lobectomy and wedge resection for the treatment can achieve satisfactory short-term outcomes.

Keywords: Congenital pulmonary airway malformation (CPAM); congenital cystic adenomatoid malformation; adult; thoracic surgery

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Introduction

Congenital lung malformations (CLMs) refer to a series of innate pulmonary malformations. Of these, congenital pulmonary airway malformations (CPAMs) are the most common. Due to the widespread use of fetal ultrasound screening, most lesions are detected prenatally as abnormal echogenic masses in the lungs. Severe fetal hydrops or pulmonary dysplasia may result in intrauterine death or neonatal respiratory distress requiring fetal intervention or emergency neonatal surgery; most live births are asymptomatic. Approximately 25% of asymptomatic patients go on to later develop symptoms, such as respiratory infections including pneumonia or a pneumothorax (1). CPAMs show malignant potential if not resected promptly. As previously reported, CPAMs can transform into a pleuropulmonary blastoma (PPB), bronchioloalveolar carcinoma (BAC), or mucinous adenocarcinoma (2-4).

Given that over 90% of patients are diagnosed before they reach the age of two years, adult CPAMs are extremely rare, and existing literature is primarily limited to case reports of several patients. Some patients are asymptomatic, while others may present with recurrent lung infections, productive cough, pneumothorax, hemoptysis, or chest pain (5-7).

Due to the rarity of the disease in adult thoracic surgery and the insufficiency of clinical workups, patients are often misdiagnosed with other pulmonary diseases. In this study, we present a series of eighteen adult CPAM cases and describe their clinical features to help clinicians better identify and manage the disease. We present this article in accordance with the STROBE reporting checklist (available at https://jtd.amegroups.com/article/view/10.21037/jtd-23-1960/rc).

Highlight box

Key findings

- Congenital pulmonary airway malformations (CPAMs) in adults show a complex presentation in terms of clinical symptoms, imaging performance and pathological findings. Thoracoscopic lobectomy and wedge resection are both feasible for adult CPAMs.

What is known and what is new?

- Adult CPAMs are extremely rare, and existing literature is primarily limited to case reports of several patients. Clinicians often fail to correctly diagnose this problem.
- Half of the CPAM patients were detected with small airway dysfunction preoperatively. We suggest that thoracoscopic surgery can achieve satisfactory short-term outcomes. The approach of wedge resection is suitable for application in older patients.

What is the implication, and what should change now?

- Adulthood CPAM has a high risk of symptoms and thoracoscopic surgery is needed to confirm the pathological diagnoses.

Methods

Patients

This study was performed in accordance with the Declaration of Helsinki (as revised in 2013). This study was approved by the Ethics Committee of China Medical University (No. 2018PS391K). All participants provided written informed consent to participate in this study. We retrospectively collected the data of 18 adults with CPAMs admitted to the Thoracic Surgery Department of Shengjing Hospital of China Medical University between 2013 and 2023. Patients were included if they underwent surgical management and had a CPAM confirmed by pathology. All the patients were treated with thoracoscopic approach, which were categorized into groups of lobectomy and wedge resection.

Data collection

Data collected included age, gender, lesion location, clinical history, preoperative diagnoses, pulmonary function test (PFT) results, treatment, surgical outcome, pathology, and postoperative complications. Preoperative chest computed tomography (CT) was performed on all CPAM patients and was evaluated by specialists in thoracic radiology and surgery. PFTs were performed for patients. We mainly documented forced vital capacity (FVC), forced expiratory volume in 1 s (FEV_1), FEV_1 to FVC ratio (FEV_1/FVC), forced expiratory flow $(FEF)_{25–75\%}$, $FEF_{50\%}$ and $FEF_{75\%}$. Pulmonary function (PF) was considered abnormal when FVC, FEV_1 and FEV_1/FVC results was below 80% predicted. Small airway function (SAF) was considered impaired when $FEF_{25–75\%}$, $FEF_{50\%}$ and $FEF_{75\%}$ results were below 70%. All patients were treated surgically by video-assisted thoracic surgery (VATS). Based on the size of lesion and the extent of involved lobes, surgical methods included wedge resection and lobectomy. Chest X-ray was reviewed 1 week and one month after surgery; chest CT was reviewed three months and one year respectively after surgery. We assessed the results descriptively as a case series.

Table 1 Baseline and preoperative characteristics of 18 patients with CPAM

Characteristics	Value (n=18)
Lesion location	
LUL	1 (5.6)
LLL	8 (44.4)
RUL	2 (11.1)
RLL	7 (38.9)
Clinical history	
None	5 (27.8)
Hemoptysis	1 (5.6)
Chest pain	1 (5.6)
Chest congestion	1 (5.6)
Productive cough	4 (22.2)
Cough + chest pain	2 (11.1)
Cough + chest congestion or dyspnea	4 (22.2)
Preoperative diagnosis	
Pulmonary infection	1 (5.6)
Pneumatocele	1 (5.6)
Lung nodule	2 (11.1)
Lung tumor	5 (27.8)
Lung tuberculosis	1 (5.6)
Bronchiectasis	4 (22.2)
Neurogenic tumor	1 (5.6)
Bronchial cyst	1 (5.6)
Abnormal lung development	1 (5.6)
CPAM	1 (5.6)

The data were expressed as n (%). CPAM, congenital pulmonary airway malformation; LUL, left upper lobe; LLL, left lower lobe; RUL, right upper lobe; RLL, right lower lobe.

Statistical analysis

The data were analyzed using IBM SPSS Statistics 25 (Armonk, NY, USA). Continuous variables were analyzed using *t*-test or Mann-Whitney *U* test. Categorical variables were compared using the Chi-squared test or Fisher's exact test. P<0.05 was considered as significant.

Results

Preoperative characteristics

Table 1 shows the preoperative characteristics of the enrolled patients. There were 4 men and 14 women in our study. The mean age at diagnosis was 54.6±16.8 years. There were 44.4% of the lesions located at the left lower lobe (LLL) and 38.9% at the right lower lobe (RLL). Only three lesions were located at the upper lobes. Five patients had no symptoms and their abnormality was detected by chance during an unrelated examination. The major presenting symptom was cough (n=10); of these 10 patients, two had intermittent chest pain on the affected side, four had recurrent pneumonia with productive cough, and four had chest congestion or dyspnea following exertion. In addition, one patient suffered from dull chest pain. Hemoptysis was reported by one patient.

CPAM lesions showed a variety of CT characteristics, including multiple cysts, nodules, ground-glass opacities, round mass, cavitation, localized inflammation, and cystic bronchiectasis as shown in *Figure 1*. Based on the radiological manifestations, the initial diagnoses were also diverse. There were 27.8% of the lesions diagnosed with lung tumor, 22.2% as bronchiectasis, 11.1% as lung nodules. Other diagnoses included pulmonary infection, tuberculosis, bronchial cyst, and neurogenic tumor. One case was diagnosed as abnormal lung development with bronchial atresia and one CPAM was correctly identified before surgery.

Table 2 shows the PFT results of our patients before surgery. The mean FVC was 95.7%±13.3% predicted, the mean FEV_1 was 88.0%±14.8% predicted, the mean FEV_1/FVC was 92.4%±12.0% predicted. Six (33.3%) patients had FEV_1 lower than 80% predicted, one patient had FVC lower than 80% predicted and two patients had FEV_1/FVC lower than 80% predicted. The mean $FEF_{25–75\%}$ was 65.2%±28.6% predicted with 11 patients less than 0.7, the mean $FEF_{50\%}$ was 68.8%±28.0% predicted with 8 patients less than 0.7, the mean $FEF_{75\%}$ was 61.6%±30.0% predicted with 12 patients less than 0.7.

Operative data

Table 3 shows the operative characteristics of the patients. All received VATS with nine wedge resections and nine

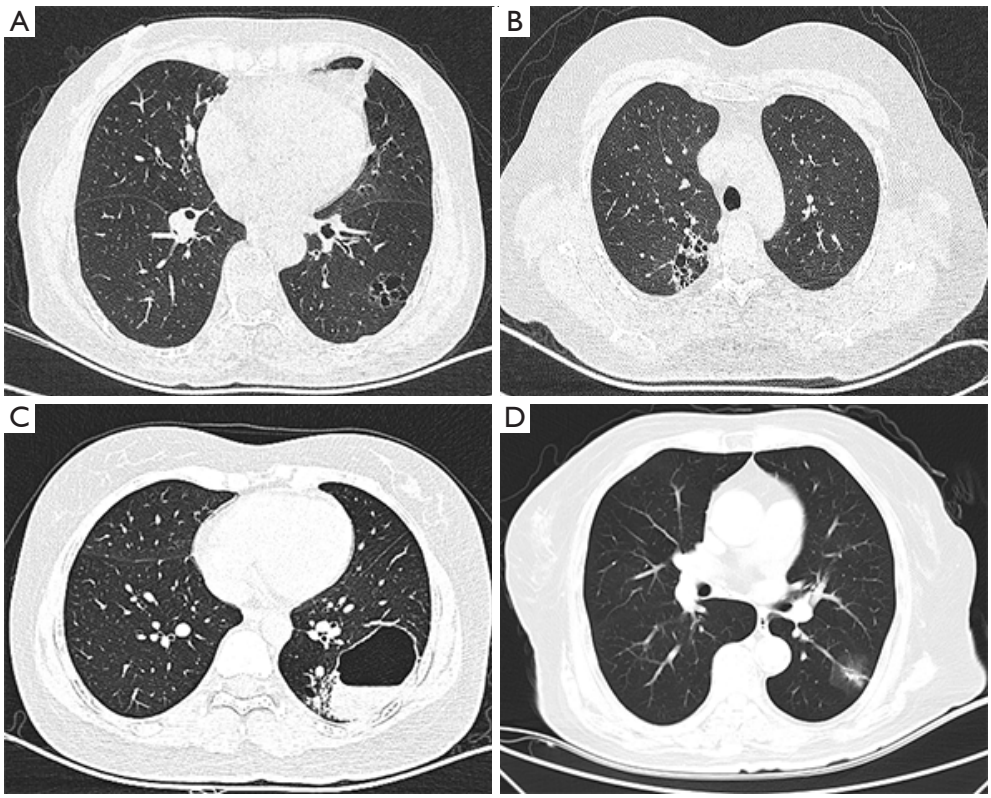

Figure 1 Appearance of congenital pulmonary airway malformations on the chest CT scans of different cases. (A) CT scan of a 66-year-old female with multiple thin-walled cysts in the left lower lung and initially diagnosed as cystic lung tumor. (B) CT scan of a 57-year-old female with cystic bronchiectasis and inflammation lesions in the right upper lobe and initially diagnosed as bronchiectasis. (C) CT scan of a 25-year-old female as a macrocystic lesion with air-fluid level in the LLL and initially diagnosed as pneumatocele. (D) CT scan of an 81-year-old female as ground-glass opacity with cavitation in the LLL and initially diagnosed as early atypical lung tumor. CT, computed tomography; LLL, left lower lobe.

Table 2 Pulmonary function test results of adult patients before surgery

PFT	Value, mean ± SD [range]
FVC (% predicted)	95.7±13.3 [71.5–123.9]
FEV_1 (% predicted)	88.0±14.8 [60.5–109.2]
FEV_1/FVC (% predicted)	92.4±12.0 [67.6–115.3]
$FEF_{25–75\%}$ (% predicted)	65.2±28.6 [25.4–126.2]
$FEF_{50\%}$ (% predicted)	68.8±28.0 [20.5–117.0]
$FEF_{75\%}$ (% predicted)	61.6±30.0 [23.3–141.1]

PFT, pulmonary function test; SD, standard deviation; FVC, forced vital capacity; FEV_1, forced expiratory volume in 1 s; FEF, forced expiratory flow.

lobectomies. There were differences in the surgical procedures received by patients of different ages, the mean age for patients under wedge resection was 64.9±3.1 years and patients under lobectomy was 44.3±5.5 years (P<0.05). Preoperative diagnosis of benign or malignant lesions was not statistically different between the two groups (P=0.62). The preoperative PF results were categorized into three grades of normal, small airway dysfunction and ventilation dysfunction, with 1/3 of patients in each grade. Whereas no statistical significance was found in the preoperative PFT results between two groups (P=0.84).

The mean operative time was 144.9±47.9 minutes. The median estimated blood loss was 50 mL [interquartile

Table 3 Comparison of operative characteristics in patients with CPAM under different surgical methods

Characteristics	All (n=18)	Wedge resection (n=9)	Lobectomy (n=9)	P value
Male gender	4 (22.2)	2 (22.2)	2 (22.2)	>0.99
Left side	9 (50.0)	5 (55.6)	4 (44.4)	>0.99
Age (years)	54.6±16.8	64.9±3.1	44.3±5.5	0.005
Preoperative symptoms	12 (66.7)	5 (55.6)	7 (77.8)	0.62
Preoperative diagnosis				0.62
Benign	12 (66.7)	5 (55.6)	7 (77.8)	
Malignant	6 (33.3)	4 (44.4)	2 (22.2)	
Preoperative pulmonary function				0.84
Normal	6 (33.3)	3 (33.3)	3 (33.3)	
Small airway dysfunction	6 (33.3)	2 (22.2)	4 (44.4)	
Ventilation dysfunction	6 (33.3)	4 (44.4)	2 (22.2)	
Operating time (min)	144.9±47.9	126.4±45.2	163.4±45.3	0.10
Estimated blood loss (mL)	50.0 [10.0–100.0]	10.0 [10.0–77.2]	50.0 [35.0–100.0]	0.25
Duration of tube drainage (days)	5.5 [4.0–6.3]	4.0 [3.3–5.5]	6.0 [5.0–8.0]	0.06
Duration of postoperative hospitalization (days)	8.0 [6.8–9.0]	7.0 [5.5–11.0]	8.0 [7.5–9.0]	0.25
Volume of tube drainage (mL)	211.0±103.7	169.2±99.7	252.8±94.7	0.08
Pathological outcome				>0.99
CPAM (+ inflammation)	13 (72.2)	6 (66.7)	7 (77.8)	
CPAM + other lesions	5 (27.8)	3 (33.3)	2 (22.2)	
Complications				>0.99
Pneumonia	5 (27.8)	4 (44.4)	1 (11.1)	
Pneumothorax	2 (11.1)	0	2 (22.2)	
Reoccurrence	0	0	0	

The data were expressed as n (%), mean ± SD, or median [IQR]. CPAM, congenital pulmonary airway malformation; SD, standard deviation; IQR, interquartile range.

range (IQR): 10.0–100.0 mL]. The median duration of tube drainage was 5.5 days (IQR: 4.0–6.3 days). The median duration of postoperative hospitalization was 8 days (IQR: 6.8–9.0 days). The mean of tube drainage volume was 211.0±103.7 mL. Postoperative chest X-rays were performed after 1 week, with which five cases of pneumonia and two cases of pneumothorax were found. Pneumonia occurred in 44.4% of the cases after wedge resection and in 11.1% of the cases after lobectomy. Pneumothorax occurred in 22.2% of lobectomies. None of the resected lesions recurred or transformed malignant during the first year after surgery. No statistical significance was found in the surgical outcomes of the two surgical procedures.

According to the pathological classification of Stocker, the samples comprised 14 cases (77.8%) of type I CPAM, characterized by the presence of pseudostratified columnar epithelium on the cyst wall, and 4 cases (28.6%) of type II CPAM, with the cyst wall composed of columnar or cuboidal epithelium. Thirteen cases were pure CPAM lesions and chronic inflammation was accompanied in five. Bronchiectasis was seen in two CPAMs with the formation of a small abscess, one of the lesions had an associated aspergilloma in the bronchial lumen and focal neuroendocrine micro-tumor. One CPAM developed atypical epithelial, one developed a carcinoma in situ, and a granulomatous lesion was discovered in one case (*Table 4*).

Table 4 Pathological outcomes of adult CPAMs

Pathological outcome	Value, n (%)
CPAM	8 (44.4)
CPAM + chronic inflammation	5 (27.8)
CPAM + bronchiectasis	1 (5.6)
CPAM + bronchiectasis + aspergillosis	1 (5.6)
CPAM + atypical epithelial hyperplasia	1 (5.6)
CPAM + carcinoma *in situ*	1 (5.6)
CPAM + granulomatous lesion	1 (5.6)

CPAM, congenital pulmonary airway malformation.

Discussion

Cough was the main presenting symptom in over half of our adult patients; 27.8% were asymptomatically detected during chance physical examinations. Other clinical symptoms included chest pain, hemoptysis, fever, dyspnea, and recurrent pulmonary infections. Rare complications such as recurrent pneumothorax have also been reported (8,9). Adults are more likely to present with symptoms than children. This might be related to the fact that lesions in children are frequently detected prenatally and surgical interventions are mostly performed before the onset of symptoms, whereas adults are more frequently diagnosed after symptoms development. In our cases, 44.4% of the CPAM lesions were located at the LLLs and 38.9% at the RLLs. This is consistent with previous literature which indicated that lesions are more common in the lower lobes.

Given the rarity of CPAMs in adult pulmonary diseases, clinicians often fail to correctly diagnose this problem. In our study, the most common misdiagnosis was a lung tumor. The lesions appeared variously as a multicystic or cavitary mass with solid components, an irregular pulmonary nodule, or ground-glass opacities. One case presented with a significant enhancement of the soft tissue component on CT. Lung cancer can present as thin-walled cysts or cavities, most of which are adenocarcinomas (10,11). Early atypical malignant lesions cannot be excluded in pulmonary nodules and ground glass opacities. In addition, four cases were diagnosed as bronchiectasis based on preoperative CT scans, which was common among the more benign misdiagnoses. Sometimes CPAMs can manifest as congenital lobar emphysema on imaging and histology confirms them to be a type 2 CPAM (12,13). Only one cystic lesion was correctly diagnosed with CPAM in a 17-year-old female. Given the heterogenous presentation of CPAMs on radiological scans, an accurate diagnosis depends on the results of pathological examinations.

Given that adult CPAMs are associated with more symptoms and risk of malignancy, surgical resection has been recommended to define the exact properties of the masses. According to the studies of pediatric CPAM, VATS might have advantages over the open thoracotomy in terms of the length of hospital stay (14-16). However, there are no consistent protocols to instruct therapies for adults with CPAM. Few studies have compared the two surgical procedures in adult CPAM. Kwon *et al.* reported that the VATS procedure is safe and feasible for adult CPAMs (17). VATS was the first choice of treatment for our cases and showed good surgical outcomes and few postoperative complications. We explored the surgical outcomes in both groups and found no significant difference in all surgical parameters between the two groups. This suggests that either wedge resection or lobectomy is feasible in adults with CPAM. Preoperative diagnosis as well as PF results did not affect the surgical strategy decision, however, wedge resection was mostly performed in older patients, probably due to the poorer PF and anesthesia tolerance. Therefore, in cases involving older adults, we suggest surgical resection as a viable treatment option, even in the absence of symptoms.

In our study, most of the CPAM patients had normal lung ventilation function before operation. However, small-airways dysfunction was found in most of our cases, which was commonly described in patients with asthma or chronic obstructive pulmonary disease (COPD). The pathology of these lesions was typically indicated by CPAM with chronic inflammation, which may be contributing to hyperplasia, spasm and narrowing of the small airway lumen. Patients usually presented with history of chronic cough.

Aspergillosis was discovered in one of the CPAM cases, who reported hemoptysis and had more complicated pathological findings with bronchiectasis and a neuroendocrine tumor. Aspergilloma within CPAM lesions was extremely rare in previous reports and only four cases have been documented (6,18-20). All of the four patients with aspergilloma presented with coughing and hemoptysis at ages of 14 to 59 years. Among them, a 59-year-old male presented with massive hemoptysis and died after postoperative hemorrhagic shock (20).

Approximately half of our pathological characterizations were not pure CPAM lesions. Chronic inflammation was present in five CPAM lesions; all these patients had

symptoms of recurrent cough. Two of our cases showed a predisposition to malignancy, including atypical proliferation and carcinoma in situ. Fortunately, none of the lesions developed invasive carcinoma in our study. According to a recent systematic review, CLMs can progress to malignancy at any age and there is no known limit to the interval for transformation from benign lesion to malignant tumor (21). In a study of 46 adult CPAMs (7), pre-malignancies or malignancies were present in over 10% of CPAM lesions. Pleuropulmonary blastoma (PPB) is a rare malignant tumor in children arising from pleuropulmonary germ cells. Type 1 PPB is indistinguishable from CPAM on imaging, as a result of its cystic components. There is also histological overlap between type 4 CPAMs and type 1 PPBs. Feinberg and colleagues identified the clinical and radiological features of the two entities to improve diagnostic and treatment management (22). BAC can arise in pre-existing type 1 CPAMs and can occur at any age throughout life (3,23,24). Type 1 CPAMs have also been reported to develop mucinous adenocarcinomas in the neonatal, childhood, and adulthood periods (4,25,26).

The main limitation of our study was the small sample size, which might lead to inaccurate statistical results. Furthermore, due to delayed detection of the disease, our cohort was older in age. Although adult CPAMs are currently uncommon, with the prevalence of conservative management in children (27,28), there may be an increase of adult patients in the future. This change could have a potential impact on treatment strategies and introduce selection bias in the research cohort. Third, our study failed to obtain long-term follow-up data, such as postoperative lung function outcomes, which could assist in evaluating prognosis.

Few articles have summarized the clinical features of CPAM in adults previously, as most are described in the form of case reports. Instead, we summarized the baseline characteristics, PFT results and surgical outcomes from 18 patients to help thoracic surgery to better acknowledge this disease. Given our small sample size, we were unable to determine an accurate generalizable algorithm for the diagnosis and management of CPAMs. Future studies are needed to determine if long-term complications develop and if neoplasms recur.

Conclusions

Adulthood CPAM has a high risk of symptoms and its correct preoperative diagnosis is difficult, largely due to the complex non-specific radiological findings. Thoracoscopic lobectomy and wedge resection for the treatment are suggested to confirm the pathological diagnoses.

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Footnote

Reporting Checklist: The authors have completed the STROBE reporting checklist. Available at https://jtd.amegroups.com/article/view/10.21037/jtd-23-1960/rc

Data Sharing Statement: Available at https://jtd.amegroups.com/article/view/10.21037/jtd-23-1960/dss

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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. This study was performed in accordance with the Declaration of Helsinki (as revised in 2013). This study was approved by the Ethics Committee of China Medical University (No. 2018PS391K). All participants provided written informed consent to participate in this study.

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