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Original Article

Prognosis of infants with congenital pulmonary airway malformations after surgery: a short and mid-term evaluation

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Abstract: Objective: To investigate the postoperative pulmonary function, imaging descriptions and complications in infants with congenital pulmonary airway malformations (CPAM), and to examine the impact of different surgical resections on the prognosis of infants. Methods: Data of 30 infants with CPAM who underwent surgery at the department of Pediatric Surgery, Guangzhou Women and Children's Medical Center from June 2021 to June 2022 were retrospectively collected and analyzed. The pulmonary function indexes of the infants during the first month and first year after surgery were analyzed to assess prognosis. Pulmonary function data from healthy individuals at similar age were collected as a control group. Results: The post-operative short-term pulmonary function was recovered to a normal level in 26.7% cases of 30 CPAM infants, with a decrease in tidal volume (VT), ratio inspiratory time to expiratory time (TI/TE), time to peak tidal expiratory flow as a proportion of expiratory time (TPTEF/TE), volume to peak expiratory flow as a proportion of exhaled volume (VPEF/VE) and mean expiratory flow as a proportion of mean inspiratory flow (MEF/MIF) when compared to the control group (all $P<0.01$). One year after operation, 25 CPAM infants received pulmonary function tests and 52% of them had indexes at normal level. There was no statistically significant difference in results of pulmonary function test between infants who received lobectomy and those who received segmentectomy ($P>0.05$). The postoperative complication rate was 26.7%. Conclusion: Over half of CPAM infants have normalized lung function one year after operation and the choice of lobectomy and segmentectomy had no significant difference on prognosis of infants.

Keywords: Congenital pulmonary airway malformation, surgery, infants, pulmonary function

Introduction

Congenital pulmonary airway malformation (CPAM) is a developmental anomaly disease of the lower airways that was once known as congenital cystic adenomatoid malformation (CCAM). The prevalence of CPAM is about 1/7200, and the prenatal diagnosis rate is 85.7% [1, 2]. Most infants with CPAM are asymptomatic at birth, but symptoms such as fever, cough, pneumothorax and recurrent pneumonia infections may occur later in life [3]. The probability of lung infection increases over time and may have a risk of malignant transformation, which makes surgery more difficult, so early surgery is advocated [4-6].

Currently in the clinic, surgery is recommended for infants with CPAM [7-9]. With the development of anesthesia and surgical techniques, the use of thoracoscopic surgery has become more common. Depending on the extent of the resection, the procedure can be categorized as lobectomy, segmentectomy, and irregular lung resection. Thoracoscopic anatomical segmentectomy is the ideal surgical option for removing the lesion while preserving normal lung tissue within the lobes and protecting lung function. Nevertheless, non-anatomic lung preservation surgery is more likely to have residual lesions than lobectomy [10, 11].

Although structural malformations of the pulmonary airways are surgically removed, abnormal lung function may still be present. There are few studies on short- and mid-term postoperative prognostic assessment in infants with CPAM, and the choice of surgical resection

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remains controversial. Follow-up of the infants at different stages of the postoperative period is useful to know the changes in pulmonary function and the incidence of complications after surgery, and to know whether the different surgical procedures have any effect on the prognosis.

Therefore, the aim of this study was to report postoperative pulmonary function outcomes, imaging changes, and complication rates, as well as to investigate the effects of lobectomy and segmentectomy on prognostic.

Materials and methods

Patients

Data of 30 infants (<3 years old) with CPAM who underwent surgery at Guangzhou Women and Children's Medical Center from June 2021 to June 2022 were retrospectively collected. The location of the lesion was evaluated preoperatively using Computed Tomography (CT) imaging and three-dimensional reconstruction techniques. Infants with non-peripheral lesion in the lobes and limited segmental involvement received anatomic segmentectomy, and those with lesion involving all lobes or more than 3 segments received lobectomy. They were followed up and postoperative clinical and pulmonary function data were recorded. This study was approved by the Ethics Committee of Guangzhou Women and Children's Medical Center.

Inclusion criteria: (1) infants who underwent surgery at Guangzhou Women and Children's Medical Center; (2) infants who were diagnosed with CPAM on pathologic findings; (3) the surgical procedure was thoracoscopically assisted anatomic segmentectomy or lobectomy; (4) infants who had complete clinical data. Exclusion criteria: (1) infants who had respiratory disease or heart disease etc. which can lead to impaired lung function and confirmed by imaging tests; (2) infants complicated with other congenital diseases or other system diseases; (3) infants that couldn't be followed up.

Methods

Pulmonary function: Tidal breathing flow-volume curve (TBFV) is commonly used in infants to measure pulmonary function. After adequate sedation, the infant is placed in the supine position and an appropriate mask is fastened to the infant's mouth and nose without leakage. The mask is connected to the flow sensor, which receives changes in pressure flow as the infant breathes and then obtains a flow signal, from which a volume signal is obtained, thus depicting a flow-volume curve [12]. The source of the flow signal in the flow-volume curve includes not only the lower airways but also the upper airways, so it reflects functional changes in the entire respiratory system. MasterScreen™ Babybody Plethysmograph pulmonary function instrument from Jaeger in Germany was used for detection.

Parameters measured during pulmonary function test include tidal volume (VT), respiratory rate (RR), ratio inspiratory time to expiratory time (TI/TE), peak flow of tidal expiratory flow (PTEF), time to peak tidal expiratory flow as a proportion of expiratory time (TPTEF/TE), volume to peak expiratory flow as a proportion of exhaled volume (VPEF/VE), mean expiratory flow as a proportion of mean inspiratory flow (MEF/MIF).

Imaging findings: We reviewed the CT findings of infants 6 months to 1 year postoperatively to look for abnormalities such as residual malformations, tumor tissue, lung hyperinflation, and scar formation. The parents of the infants were informed about the indications and purpose of the radiologic examinations and consent was obtained. All images were reviewed by two pediatric radiologists. Pulmonary function data from healthy individuals at similar age were collected as a control group to observe the pulmonary function recovery degree of CPAM infants.

Complications: The postoperative infants were followed up on outpatient and the incidence of complications were calculated. If clinical complications occurred within 1 month after surgery, they were categorized as short-term complications such as pneumothorax, pleural effusion, bleeding, and infection. Complications occurred after one month post operation were defined as late complications. The diagnostic criteria are as follows: (1) Cough lasting more than two months within one year; (2) Lower respiratory tract infection diagnosed by a physician more than three times; (3) Required inhaled bronchodilator or antibiotic therapy; (4) Thoracic deformity.

Table 1. Patients' characteristics and outcomes

	Overall	Lobectomy	Segmentectomy
Numbers	30	9 (30%)	21 (70%)
Sex (male/female)	18/12	9/0	9/12
Preoperative symptoms	8 (26.7%)	2 (6.7%)	6 (20%)
Location of CPAM			
Left	11 (36.7%)	1 (3.3%)	10 (33.3%)
Right	19 (63.3%)	8 (26.7%)	11 (36.7%)
Pathology			
I	13 (43.3%)	3 (10%)	10 (33.3%)
II	17 (56.7%)	6 (20%)	11 (36.7%)
Age (months)	10.13±1.47	8.67±0.94	10.76±2.06
Height (cm)	72.45±1.70	72.50±1.90	72.40±2.31
Body weight (kg)	8.91±0.35	8.92±0.47	8.91±0.46

Note: CPAM, congenital pulmonary airway malformations; results are given as the number (%) or as the mean ± standard error of mean.

Statistical analysis: Data analysis was performed using IBM SPSS 25.0. Shapiro-Wilk method was used to assess the normality of the data. Measurement data conformed to a normal distribution were expressed as mean ± standard error of the mean and were analyzed by independent t-tests, and those that didn't conform to a normal distribution were expressed as median (P25, P75) and analyzed by Wilcoxon signed rank-sum test. Categorical variables were expressed as numbers and percentages and were assessed using Fisher's exact test. $P<0.05$ was considered as a statistically significant difference, and all *p*-values were two-tailed.

Results

Patient characteristics

A total of 30 infants were included in the study, including 18 boys and 12 girls with a mean weight of 8.91±0.35 kg. They all underwent thoracoscopic surgery with a mean age at surgery of 10.13±1.47 months. There were 11 cases with lesions on the left side and 19 cases with lesions on the right side, and 8 cases with preoperative symptoms of pulmonary infection. Anatomic segmentectomy was performed in 21 cases and lobectomy in 9 cases. Twenty-five of 30 underwent pulmonary function tests at one year post-operation with a mean age of 20.68±0.92 months. Nine of them underwent lobectomy and sixteen underwent segmentectomy. The data are summarized in **Table 1**.

Short-term prognosis

Pulmonary function: The short-term pulmonary function of the infants with CPAM was as follows: tidal volume decreased in 2 (6.7%) infants and the rest had normal tidal volume. 8 (26.7%) infants had normal pulmonary function results, 13 (43.3%) had mild airway obstruction, and 7 (23.3%) had moderate airway obstruction. There was no statistically significant difference in pulmonary function between the lobectomy and segmentectomy group (All $P>0.05$). As shown in **Table 2**.

Thirty healthy individuals at similar age and weight to the CPAM infants (the CPAM group) were selected as the control group, with mean age of 9.83±1.03 months, including 15 boys and 15 girls. The mean VT/kg values, TI/TE, TPTEF/TE, MEF/MIF and VPTEF/VE of the infants in the CPAM group were lower than those in the control group (all $P<0.01$). The mean PTEF in the CPAM group were higher than those in the control group ($P=0.043$). There was no statistical difference in RR between the two groups ($P=0.096$). The results of pulmonary function tests are summarized in **Table 3**.

Postoperative complications: Five of the thirty infants (16.7%) had short-term complications within 1 month of surgery. Four of the five infants underwent segmentectomy, and complications included two pleural effusions, one hemorrhage, and one bronchopneumonia. One infant underwent lobectomy and developed respiratory failure. They were treated and discharged safely.

Medium-term prognosis

Pulmonary function: Patients were followed up one year after surgery and pulmonary function tests were performed. Data of 25 children were collected. They all had normal tidal volumes and thirteen children (52%) had normal pulmonary airway function. Of the 9 patients in the lobectomy group, 6 were normal and 3 had mild airway obstruction. Pulmonary function results in the segmentectomy group were normal in

Table 2. Short and Medium-term pulmonary function test results after surgery for CPAM

Group	Short-term				Medium-term			
	Overall	Lobectomy	Segmentectomy	P	Overall	Lobectomy	Segmentectomy	P
Number	(N=30)	(N=9)	(N=21)		(N=25)	(N=9)	(N=16)	
Normal	8 (26.7%)	3 (10%)	5 (16.7%)	0.666	13 (52%)	6 (24%)	7 (28%)	0.411
Mild airway obstruction	13 (43.3%)	2 (6.7%)	11 (36.7%)	0.229	6 (24%)	3 (12%)	3 (12%)	0.630
Moderate airway obstruction	7 (23.3%)	3 (10%)	4 (13.3%)	0.640	5 (20%)	0 (0%)	5 (20%)	0.123
Severe airway obstruction	0 (0%)	0 (0%)	0 (0%)	-	0 (0%)	0 (0%)	0 (0%)	
Restrictive disease	2 (6.7%)	1 (3.3%)	1 (3.3%)	0.517	0 (0%)	0 (0%)	0 (0%)	

Note: CPAM, congenital pulmonary airway malformations; results are given as the number (%).

Table 3. Outcomes in the CPAM group vs. the control group and in the lobectomy group vs. the Segmentectomy group at one year postoperatively

Group	Short-term			Medium-term			Surgery		
	Control	CPAM	P	Control	CPAM	P	Lobectomy	Segmentectomy	P
Number	(N=30)	(N=30)		(N=18)	(N=25)		(N=9)	(N=16)	
Age (months)	9.83±1.03	10.13±1.47	0.868	21.83±1.66	20.68±0.92	0.519	20.67±1.29	20.68±0.88	0.991
VT/kg (ml/kg)	7.68±0.18	6.85±0.18	0.002	7.94±0.24	7.94±0.21	0.999	7.91±0.35	8.06±0.18	0.659
RR (1/min)	27.60 (22.15, 31.50)	29.95 (23.83, 34.50)	0.096	24.42±1.01	24.11±0.89	0.818	23.21±1.48	24.74±0.79	0.371
Ti/Te	0.68±0.16	0.61±0.13	0.002	0.62±0.02	0.61±0.02	0.553	0.60±0.26	0.61±0.17	0.802
TPTEF/TE (%)	37.59±1.73	24.56±1.46	0.000	33.96±1.89	29.16±2.00	0.101	32.26±2.92	27.36±1.83	0.203
VPTEF/VE (%)	34.80 (31.25, 42.83)	27.00 (23.58, 33.03)	0.000	34.67±1.44	30.94±1.67	0.114	32.99±2.35	29.61±1.56	0.298
PTEF (ml/s)	66.93±2.82	74.43±2.51	0.043	78.55±4.54	82.60±3.86	0.502	75.56±5.63	86.50±3.47	0.137
MEF/MIF (%)	67.45±1.60	61.20±1.34	0.004	62.79±1.71	61.02±1.73	0.484	60.67±2.46	61.22±1.65	0.872

Note: CPAM, congenital pulmonary airway malformations; VT/kg = tidal volume/kg; RR = respiratory rate; TI/TE = ratio inspiratory time to expiratory time; TPTEF/TE = time to peak tidal expiratory flow as a proportion of expiratory time; VPEF/VE = volume to peak expiratory flow as a proportion of exhaled volume; PTEF = peak flow of tidal expiratory flow; MEF/MIF = mean expiratory flow as a proportion of mean inspiratory flow. Results are given as the mean ± standard error of mean.

seven cases, mild airway obstruction occurred in three cases and moderate airway obstruction in five cases. As shown in **Table 2**.

For the medium-term evaluation, data of 18 healthy individuals at similar age and with similar weight to the group were collected as the control group, with a mean age of 21.83±1.66 months, including 9 boys and 9 girls. The pulmonary function results showed that at one-year post-operation, there was no significant difference in VT/kg (P=0.999), RR (P=0.818), Ti/Te (P=0.553), TPTEF/TE (P=0.101), VPTEF/VE (P=0.114), PTEF (P=0.502) and MEF/MIF (P=0.484) between the two groups. The results of pulmonary function tests are summarized in **Table 3**.

Comparing the pulmonary function results in the lobectomy group and segmentectomy group, there was no significant difference in VT/kg (P=0.659), RR (P=0.371), Ti/Te (P=0.802), TPTEF/TE (P=0.203), VPTEF/VE (P=0.298), PTEF (P=0.137) and MEF/MIF (P=0.872) between the two groups.

Imaging findings: One (3.3%) infant with type I CPAM had a residual cystic lesion in the operated lung suggested by a follow-up CT one year after surgery. No residual malformations or tumors were found in the other infants. Hyperinflation was seen in five children (16.7%) and almost all had scarring (mild). No chest deformities or rib fusions were found. As shown in **Figure 1**.

Complications: At one year post-operation, 25 of 30 infants had follow-up data. Among them, 3 (12%) infants developed complications, two of whom experienced three or more than three episodes of pneumonia and one of whom had chronic cough, all after segmentectomy. No complications such as chest deformity or asthma were found during the follow-up period. As shown in **Table 4**.

Discussion

CPAMs are characterized by hyperplasia and dilatation of the terminal bronchus [13, 14]. In 1949, Ch'in and Tang named CPAM as congenital cystic adenomatoid malformation (CCAM),

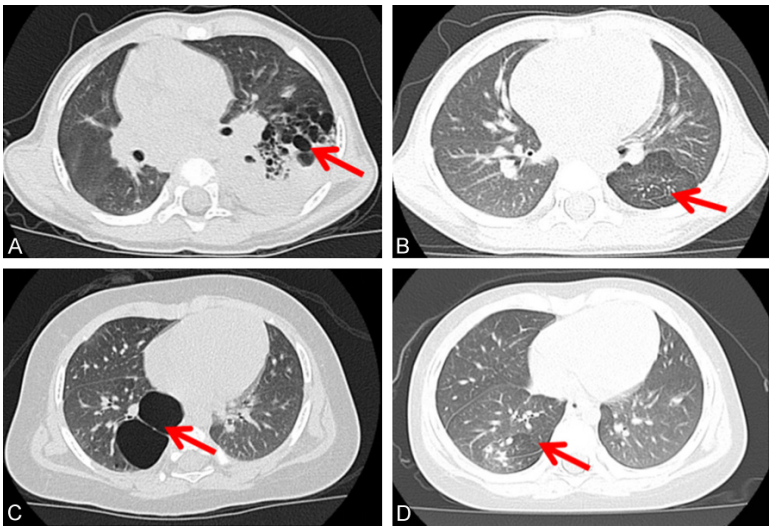

Figure 1. A: Preoperative CT of multiple cystic lesions in the left lung. B: Hyperinflation in operated lung. C and D: Preoperative CT and postoperative CT showed a few residual cystic lesions.

a rare disorder [15]. In 1977, Stocker et al. classified CCAM into types I, II, and III based on cyst size and pathologic features [16]. In 2002, Stocker renamed it as congenital pulmonary airway malformation (CPAM) based on the site of origin, pathologic features of the lesion and clinical characteristics, and subtyped it into 0, I, II, III and IV. Among them, type I accounts for 60-70% of cases, which is a single or several thick-walled larger cysts (the diameter of the cysts is about 2-10 cm); type II accounts for 10% to 15% of cases and consists of multiple small, evenly distributed cysts and the diameter of the cysts is about 0.5 to 2 cm [13].

The effect of lung tissue resection on pulmonary function is a continuing concern for pediatric thoracic surgeons. It has been shown that lung volume decreases immediately after lobectomy, as well as TPTEF/TE, suggesting small airway obstruction [17, 18]. In our study, we found that pulmonary function results in CPAM infants 1 month after surgery showed lower tidal volumes than normal infants, as well as decreased TI/TE, TPTEF/TE, VPTEF/VE, and MEF/MIF, suggesting airway obstruction and airflow limitation [19, 20]. We also found that 66.6% of CPAM infants showed airway obstruction and 6.7% showed a decrease in tidal volume one month after surgery, which may be associated with damage to lung tissue from a primary lesion, a history of recurrent preoperative pneumonia infections, or surgical removal of lung tissue.

In infancy, compensatory lung growth may occur after partial lung resection, and most studies have used lung function as an important indicator of lung growth. In 1980, McBride et al. followed children who underwent lobectomy in infancy, and found that the total lung volume was nearly normalized, despite the fact that 8-45% of the lung tissue had been removed [21]. In 1982, Frenckner and Freyschuss followed patients undergoing lobectomy for up to 11 years and showed that the total lung volume was approximately 90% of the predicted normal value, suggesting compensatory growth of the remaining lung tissue [22]. These are consistent with Mandaliya's findings [23]. However, this does not mean that pulmonary function is normal in compensatory growing lung tissue. Elevated residual volume, local ventilation abnormalities, and decreased perfusion after lobectomy suggest that expansion of the remaining lung may be volume compensation for tissue loss, but it may not be functional lung tissue. It has been shown that anatomical segmentectomy preserves more normal lung tissue than lobectomy, which is more conducive to recovery of lung function [18]. However, the findings of this study do not support this view. In terms of postoperative complications, we found that infants in the lobectomy group recovered well and had a lower incidence of short-term and late complications.

The anatomical landmarks of lobectomy are more obvious than those of segmentectomy making the operation relatively simple, and the surgeons needn't to find the demarcation line between normal and diseased lung segments during the operation which makes the operation time relatively shorter, so the probability of complications such as postoperative hemorrhage, infection, and pneumothorax is lower.

From this perspective and the results of our study, the tendency is to choose lobectomy for surgical treatment. However, some studies have found that there is no significant difference between the duration of surgery, hospital-

Table 4. Complication after surgery for CPAM

Group	Short-term				Medium-term			
	Overall	Lobectomy	Segmentectomy	P	Overall	Lobectomy	Segmentectomy	P
Numbers	30	9	21		25	9	16	
Complications	5 (16.67%)	1 (11.11%)	4 (19.05%)	1.000	3 (12.00%)	0 (0%)	3 (18.75%)	0.534
Pleural effusion	2 (6.67%)	0 (0%)	2 (9.52%)	-	0 (0%)	0 (0%)	0 (0%)	-
Bleeding	1 (3.33%)	0 (0%)	1 (4.76%)	-	0 (0%)	0 (0%)	0 (0%)	-
Infection	0 (0%)	0 (0%)	0 (0%)	-	0 (0%)	0 (0%)	0 (0%)	-
Bronchopneumonia	1 (3.33%)	0 (0%)	1 (4.76%)	-	0 (0%)	0 (0%)	0 (0%)	-
Respiratory failure	1 (3.33%)	1 (11.11%)	0 (0%)	-	0 (0%)	0 (0%)	0 (0%)	-
Have three or more pneumonias after discharge	0 (0%)	0 (0%)	0 (0%)	-	2 (8.00%)	0 (0%)	2 (12.50%)	-
Asthma after discharge	0 (0%)	0 (0%)	0 (0%)	-	0 (0%)	0 (0%)	0 (0%)	-
Persistent cough	0 (0%)	0 (0%)	0 (0%)	-	1 (4.00%)	0 (0%)	1 (6.25%)	-
Thoracic deformity	0 (0%)	0 (0%)	0 (0%)	-	0 (0%)	0 (0%)	0 (0%)	-

Note: CPAM, congenital pulmonary airway malformations; results are given as the number (%).

ization, and the choice of lobectomy or segmentectomy on postoperative complications, and that the age at surgery and the presence of preoperative infection are important risk factors [14, 24, 25]. In any case, the choice between lobectomy and segmentectomy also depends on the extent of the lesion and the technical experience of the surgeon. Anatomic segmental resection requires not only fine surgical skills on the part of the surgeon, but also familiarity with local vascular and bronchial anatomy.

There were several limitations to this study. The first limitation of our study was the limited sample size. Secondly, the rate of lost visits at one year post-operation was 16.7%. This may have affected the results of the postoperative examination. The third limitation was the shortcomings of the experimental design resulting in limited objective reference indices. Examination such as respiratory dynamics and functional residual volume could have been performed to improve the validity of the results. Finally, the follow-up period in this study was short. Therefore, a multicenter and randomized study with larger cohort of patients will be performed in the further with longer time follow up and more significant data analysis to assess the long-term prognostic outcomes of children with CPAM.

In conclusion, short-term pulmonary function (1 month after surgery) in infants with CPAM was abnormal, suggesting small airway obstruction. At one year post-operation, their pulmonary function returned to normal level. The choice of lobectomy or segmentectomy in infancy has little effect on their prognosis. The incidence of postoperative complications was low, but there may be a risk of residual lesions or emphysema.

Disclosure of conflict of interest

None.

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