Surgical Outcomes for Congenital Lung Malformations: 10 Years Experience at a Single Center

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Objective: *To study the surgical outcome of congenital lung malformation and natural history of the disease at Srinagarind Hospital.*

Material and Method: *Retrospectively review the medical records of 25 patients diagnosed with congenital lung malformation who underwent surgical treatment between January 2001 and December 2011.*

Results: *Twenty-five patients diagnosed with congenital lung malformation underwent surgery: 16 males (64%), 9 females (36%), median age seven months, median body weight 6 kg. Twelve (48%) had congenital cystic adenomatoid malformation congenital cystic adenomatoidmaflormation (CCAM), seven (28%) pulmonary sequestration, four (16%) congenital lobar emphysema, and one a bronchogenic cyst (4%). The most common presenting symptoms were respiratory tract infection (14, 56%), respiratory distress (7, 28%), lung abscess (1, 4%), hemoptysis (1, 4%), and asymptomatic (2, 8%). Post-operative mortality over 30 days was 0. Complications included lung infection, atelectasis, post-operative bleeding, and wound infection. Post-operative follow-up (lasting between 1-8 years) revealed normal tolerance to daily activity without any respiratory problems.*

Conclusion: *Surgical treatment for congenital lung malformations had a good result with few complications.*

Keywords*: Congenital lung malformation, Congenital bronchopulmonary malformation, Surgical outcome, Congenital cystic adenomatoid malformation (CCAM), Congenital pulmonary airway malformation (CPAM), Congenital lobar emphysema, Pulmonary sequestration, Bronchogenic cyst, Post-operative complications*

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Congenital lung malformation diseases are uncommon. Symptoms vary: the patients may not have any or develop dyspnea requiring mechanical ventilation. The most common disease is congenital cystic adenomatoid malformation (CCAM), sometimes referred to as congenital pulmonary airway malformation (CPAM). The incidence of this disease is between 1:10,000 and 1:35,000[11], followed by congenital lung cyst and pulmonary sequestration, respectively. The disease can be detected by ultrasound prenatally—the most common lesion seen being cystic lesions[2]—so the doctor can make a treatment plan before delivery. Surgery is the current treatment approach if a patient has respiratory complications during the neonatal period; however, in patients without any symptoms, most physicians prefer watchful waiting rather than surgery[3]. In some patients, complications such as recurrent respiratory tract infections necessitate surgery. Thus, the approach depends on the symptoms. The results of surgical treatment are usually satisfactory—in particular, resection in younger patients as the lung tissue parenchyma can regenerate and regain normal function.

In Thailand there is no collective data about these diseases therefore the purpose of the present study was to review the surgical outcome and natural history of congenital lung disease among Thai patients.

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Material and Method

Patient population

The medical records of patients diagnosed with a congenital lung malformation and operated upon were retrospectively reviewed between January 2001 and December 2011. The data includes the prenatal period (including gestational age when anomalies were detected, abnormal imaging detected by ultrasound) and the postnatal period (including age, sex, weight,

diagnosis based on signs and symptoms, abnormal imaging of chest X-ray or CT scan). The operative data (viz., surgical procedure), post-operative complications, duration required for mechanical ventilation, duration of chest drain and the pathological report confirm the diagnosis were collected.

Ethical issues and statistical methods

The present study was performed at Srinagarind Hospital, Faculty of Medicine, Khon Kaen University. The study was approved by the ethical board committee of the university. The descriptive statistics are used as frequency and percentage for categorical variables, mean (SD) or median (range) for continuous variables as appropriate.

Results

Of the 25 patients,16 were males (64%): the female to male ratio was 1:1.6 (Table 1). The age range was between one day and 30 years: median seven months. Fifteen patients (60%) were under one year of age. The median weight was 6 kg (range, 2.3 to 53.9 kg).

Five patients (20%) were diagnosed during the prenatal period by ultrasound, which revealed characteristic multiple cystic lesions. There was no relationship between the disease and any other congenital disorder(s) during pregnancy (i.e., hydropfetalis, fetal ascites, or fetal pleural effusion). CCAM was diagnosed in 12 (48%) of the 25 patients (Table 2), pulmonary sequestration in seven (28%),

Table 1. Overview data of 25 patients diagnosed with congenital pulmonary malformation

Patient	Sex	Age	BW (kg)	Diagnosis	Operation
1	F	1 d	2.9	LUL, CCAM	LUL lobectomy
2	F	1 d	3.1	RLL, CLE	Right pneumonectomy
3	M	1 d	2.3	RML bronchogenic cyst	RML lobectomy
4	M	4 d	3.6	RLL, CCAM	RLL lobectomy
5	M	29 d	3.9	LUL, CLE	LUL lobectomy
6	M	1 m	2.5	RLL, CLE	Right pneumonectomy
7	F	1 m	3.2	RLL, CLE	RLL lobectomy
8	F	1 m	3.1	RLL, CCAM	RLL lobectomy
9	M	2 m	2.7	RLL, CCAM	Right bilobectomy
10	M	4 m	6.0	RLL, CCAM	RLL lobectomy
11	M	5 m	3.9	Right primary bronchus hypoplasia	Right pneumonectomy
12	M	7 m	5.9	RLL, CCAM	RLL lobectomy
13	M	9 m	6.0	RLL, CCAM	RLL lobectomy
14	M	11 m	6.3	LLL extrapulmonary sequestration	LLL lobectomy
15	F	2 y	10.0	LLL intrapulmonary sequestration	LLL lobectomy
16	M	2 y	10.0	RLL, CCAM	RLL lobectomy
17	M	10 m	8.2	LLL extrapulmonary sequestration	LLL lobectomy
18	M	4 y	12.0	RLL, CCAM	RLL lobectomy
19	F	5 y	15.0	RLL, CCAM	RLL lobectomy
20	F	7 y	20.0	RLL, CCAM	RLL lobectomy
21	M	7 y	19.0	LLL intrapulmonary sequestration	LLL lobectomy
22	F	9 y	27.0	LLL, CCAM	LLL lobectomy
23	M	22 y	53.9	LLL extrapulmonary sequestration	LLL lobectomy
24	M	30 y	50.0	RLL intrapulmonary sequestration	Bilobectomy
25	F	30 y	53.0	LLL extrapulmonary sequestration	LLL lobectomy

M = male; F = female; d = day; m = month; y = year; BW = body weight; CCAM = congenital cystic adenomatoid malformation; CLE = congenital lobar emphysema; RUL = right upper lobe; RML = right middle lobe; RLL = right lower lobe; LUL = left upper lobe, LLL = left lower lobe

congenital lobar emphysema in four (16%), and bronchogenic cyst in one (4%).

Almost all of the patients had presenting symptoms before admission; only two cases were asymptomatic and detected by a physician during a checkup. The manifestation of symptoms usually occurred in the first year of life (15; 60%), between 1 and 5 years (4; 16%), between 5 and 10 years (3; 12%), and older than 10 years (3; 12%).

The presenting symptoms included respiratory tract infection (14; 56%), respiratory distress (7; 28%), lung abscess (1; 4%), hemoptysis (1; 4%), and asymptomatic (2; 8%) (Table 4). The most common recurrent infection symptom was CCAM although patients who had presenting symptoms with respiratory distress were usually found to have congenital lobar emphysema.

All of the patients were examined using preoperative CXR, while a pre-operative CT scan was performed in 20 (80%). The most common image finding from CT was cystic lesions.

Since most of the patients had symptoms at admission, all of them might be classified urgent cases: three needed emergency surgery, 18 urgent surgery, and four elective surgery. Seven patients required pre-operative intubation and ventilator support. All of

Table 2. Demographic data of patients with CCAM, CLE, pulmonary sequestration, bronchogenic cyst and bronchopulmonary dysplasia included preoperative status

	CCAM (n = 12)	CLE (n = 4)	Pulmonary sequestration (n = 7)	Bronchogenic cyst (n = 1)	Primary bronchus hypoplasia (n = 1)
Sex					
Male	7 (28%)	2 (8%)	5 (20%)	1 (4%)	1 (4%)
Female	5 (20%)	2 (8%)	2 (8%)	0	0
Age	7 m (1 d-9 y)*	29 d (1 d-1 m)*	7 y (11 m-30 y)*	1 d (GA 35 week)	5 m
BW (kg)	6 (2.7-36)*	3.2 (2.5-3.9)*	19 (3.6-53.9)*	2.3	3.9
Prenatal diagnosis	6 (24%)	1 (4%)	-	-	-
Pre-operative ventilator support	4 (16%)	1 (4%)	-	-	-

* Data present as number or median (percent or range)
d = day; m = month; y = year; BW = body weight; CCAM = congenital cystic adenomatoid malformation; CLE = congenital lobar emphysema; GA = gestational age

Table 3. Diagnosis and distribution of lesions

	Right	No.	Total right lesion (n = 16)	Left	No.	Total left lesion (n = 9)	Total (n = 25)
CCAM	RUL	0 (0%)	10 (40%)	LUL	1 (4%)	2 (8%)	12 (48%)
	RML	-		LLL	1 (4%)		
	RLL	10 (40%)					
Pulmonary sequestration	RUL	-	1 (4%)	LUL	-	6 (24%)	7 (28%)
	RML	-		LLL	6 (24%)		
	RLL	1 (4%)					
CLE	RUL	-	3 (12%)	LUL	1 (4%)	1 (4%)	4 (16%)
	RML	-		LLL	-		
	RLL	3 (12%)					
Bronchogenic cyst	RUL	-	1 (4%)	LUL	-	0	1 (4%)
	RML	1 (4%)		LLL	-		
	RLL	-					
Bronchus hypoplasia	Right lung	1 (4%)	1 (4%)	-	-	0	1 (4%)

data present as number (percent)
CCAM = congenital cystic adenomatoid malformation; CLE = congenital lobar emphy sema; RUL = right upper lobe; RML = right middle lobe; RLL = right lower lobe; LUL = left upper lobe; LLL = left lower lobe

Table 4. Manifesting symptoms and diseases

Clinical manifestation	Diagnosis	No.	Total (%)
Recurrent pneumonia	CCAM	9	14 (56)
	Pulmonary sequestration	4	
	Bronchogenic cyst	1	
Respiratory distress	CLE	4	7 (28)
	CCAM	1	
	Bronchus hypoplasia	2	
Lung abscess hemoptysis	CCAM	1	1 (4)
	Pulmonary sequestration	1	1 (4)
No symptom	Pulmonary sequestration	2	2 (8)

Table 5. Type of Operative procedures correlated with affected lesion

Operation	Diagnosis	No.	Total (%)
RLL lobectomy	CCAM	9	10 (40)
	CLE	1	
LLL lobectomy	Extrapulmonary sequestration	4	7 (28)
	Intrapulmonary sequestration	2	
	CCAM	1	
Right pneumonectomy	CLE	2	3 (12)
	Bronchopulmonary hypoplasia	1	
Right bilobectomy	CCAM	1	2 (8)
	Intrapulmonary sequestration	1	
LUL lobectomy	CCAM	1	2 (8)
	CLE	1	
RML lobectomy	Bronchogenic cyst	1	1 (4)

the patients underwent resection of the affected lobe. The type of surgery included: RLL lobectomy (10; 40%) LLL lobectomy, (7; 28%), right pneumonectomy (3; 12%), right bilobectomy (2; 8%), LUL lobectomy (2; 8%), and RML lobectomy (1; 4%) (Table 5).

After surgery, the patients were extubated on average 2.6 days and moved out of the ICU. Post-operative complications consisted of lung infection, atelectasis, post-operative bleeding requiring surgery and wound infection (Table 6). There was no post-operative mortality within 30 days of surgery, albeit one patient died after surgery due to sepsis and with multi-organ failure about two months later.

All of the tissue specimens were sent for pathological examination to confirm.

The range of hospital stay was five to 81 days (mean, 21 days). All of them were followed-up at hospital between one and nine years after discharge. All of them were well developed, had normal daily activities with no restriction, and none had any respiratory problems or recurrence of disease.

Table 6. Post operative complications

Complication	Number (%)
Atelectasis	1 (4)
Pneumonia	1 (4)
Hemorrhage	1 (4)
Wound infection	1 (4)

Discussion

Congenital bronchopulmonary malformations are uncommon diseases. Most of the abnormalities are classified as cystic lung lesions. The pathology of disease begins during embryonic development. Normally, the lower respiratory system will generate from the laryngotracheal diverticulum within four weeks of gestation[21] and development of the lung buds at GA five weeks. Over the next 24 weeks, all 17 branches will develop. During this period of development, many factors lead to the abnormal developments. It is hypothesized that the major causes of airway obstruction[21] are cystic changes arising in the lung parenchyma.

It can be difficult to make a definitive diagnosis because congenital lung abnormalities often have similar lesions among the various presentations of disease. The chest X-ray findings are similar such as cystic lesions from CCAM (congenital cystic adenomatoidmal formations) or CLE (congenital lobar emphysema). Consequently, a CT scan or MRI and pathological results help to confirm and make a final diagnosis.

The authors found that patients diagnosed with congenital lung malformations were predominately male, as was found in other studies[4,6,10]. Most of our patients came to hospital with presenting symptoms and 60% had presenting symptoms within the first year of life. The most common manifestation was respiratory tract infection, which was usually found in CCAM. The second most common symptom was respiratory distress found mainly among patients with CLE patients. Two asymptomatic patients, both incidental findings diagnosed with pulmonary sequestration.

The most common diagnosis in congenital lung malformation patients was CCAM, also currently called CPAM (congenital pulmonary airway malformation) followed by pulmonary sequestration (Table 3, 4). CCAM (or CPAM) is one of the most common diseases for persons with cystic lung lesions. In the present study, of the 25 patients, 11 (44%) were diagnosed with CCAM, a similar proportion as found in other studies[3]. The disease can be classified into four types based on Stocker's Classification[20]. The most common presenting symptom in older children is respiratory infection, while respiratory distress is usually found in neonates. Although the older children usually come to the hospital with symptoms of respiratory tract infection or pneumothorax[8], some may be asymptomatic. In the present study, we found that most lesions involved the right upper lobe.

The most common manifestation in CLE (or congenital lobar emphysema) is respiratory distress from over-inflation of the lung tissue caused by collapsing of the bronchus leading to air-block hyperinflation[10] or caused by overgrowth of the alveoli. An increased number of alveoli also cause hyperinflation. In present study, the authors found that all CLE patients had respiratory distress and the symptoms in neonates usually presented as an emergency needing surgical treatment. Two of three cases underwent emergency surgery and the most commonly found lesion was at the RUL in contrast to the usual LUL found in other studies[21].

Pulmonary sequestration is an abnormality arising from accessories of the lung buds during the early stages of embryonic development and usually occurs in normal lung tissue. If sequestration occurs during later development, it will cause an extra pulmonary sequestration. In present study, the authors found seven patients diagnosed with this disease; four of whom had symptoms of respiratory tract infection. All four had symptoms manifest after the neonatal period (between 2 and 30 years) and one came to see the doctor because of hemoptysis, while two patients were asymptomatic. The lesions were most commonly found at the left lower lobe but in one patient at the right lower lobe, consistent with previous studies[21].

One patient was diagnosed with bronchogenic cyst and had pulmonary infection symptoms, similar to other patients diagnosed with lung cyst (s), and came to hospital with infected lung cysts that progressed to a lung abscess. The last patient was diagnosed with congenital bronchial hypoplasia that developed into respiratory distress, hence need admission.

Even though congenital lung malformation disease can be detected during early pregnancy (i.e., a prenatal diagnosis), the authors found only five cases (20%) detected by ultrasound and diagnosed during the prenatal period and all five had persistent lung lesions until delivery. In fact, in the previous decade, people in the northeastern Thailand rarely came to hospital for ANC (antinatal care) even though the authors had a well-developed ANC system. This major shortcoming in Thailand's Public Health System was addressed, resulting in more early diagnosis of congenital lung abnormalities so a pre-delivery plan was set to improve treatment outcomes.

All of our patients underwent chest X-ray, but lesions were seen in only five (20%), all of which were cystic lesions diagnosed as CCAM. Among the patients diagnosed with CLE, the chest X-ray primarily indicated characteristic hyperinflation in two cases. In the two patients diagnosed with pulmonary sequestration, the chest X-ray showed a lesion-like lung mass. Chest X-ray has a low sensitivity and specificity for diagnosing diseases with cystic lesions. CT scan and MRI are considered important adjuncts for diagnosis because the lesions of each disease have specific characteristics that can be identified better on CT scan or MRI[24]. In present study, all of the patients were examined by CT scan before surgery, but there were some limitations because the appearance of the cystic lesions were somewhat similar among diseases and could not be definitively

differentiated. Therefore, tissue pathology is needed to confirm the diagnosis.

Since most of the patients had presenting symptoms at admission and even though two were asymptomatic, the imaging could not rule out tumors, so all of the patients were treated by surgery. Three patients underwent emergency pneumonectomy due to respiratory distress; two of these were diagnosed as CLE, and one with bronchopulmonary dysplasia. The standard surgical technique was used including open thoracotomy with resection taking approximately two hours. There were no complications. After surgery, the patients were all transferred to the intensive care unit. The approximate intubation time was 2.6 days.

There are two approaches for asymptomatic patients. Some physicians think postponing surgery is prudent while others argue that early surgery is low risk with good results even without symptoms[15]. Since almost all of our patients had symptoms upon admission and needed operative treatment, it is difficult to decide which approach might be better. In our opinion, if the physician decides to delay surgery, he should closely monitor the patient for early detection of any abnormalities in order to prevent potentially serious complications. Further study is needed to confirm the benefits of either approach.

The current trend in surgical treatment is a minimally-invasive approach even for congenital lung malformations. Several studies have compared the advantages of minimally invasive procedures with conventional techniques or open chest surgery. The results indicate no significant statistically difference in safety, outcomes and post-operative complications. A minimally-invasive approach is better in terms of post-operative recovery, less pain and a shorter hospital stay[17].

There was no post-operative mortality within 30 days of surgery but one patient died due to multiple medical problems included sepsis and multi-organ failure two months after surgery. Other complications included wound infection, atelectasis, lung infection, and post-operative bleeding from surgical technique that required re-operative treatment (Table 6).

After surgery, all of the patients underwent follow-up between one and nine years. All had good quality of life, tolerated normal daily activity, and had no recurrence of the disease. Surgical treatment was confirmed as the treatment of choice for congenital lung diseases[2,6,10].

Conclusion

Based on this retrospective review, we conclude that surgical treatment is the treatment of choice because it provides a good outcome with few complications. The key to successful management for congenital lung malformation is early diagnosis in order to prevent the various complications that put the patient at risk of pulmonary problems. Further study is needed to clarify the controversial issues of postponement of surgery in asymptomatic patients and the advantage of a minimally invasive approach.

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What is already known on this topic?

Congenital lung malformations are rare diseases. There is no obvious guideline for treatment but surgery is acceptable for standard treatment and has been reported for good results in other countries. However, current trends of treatment may observe the clinical in asymptomatic patients rather than perform immediately surgery.

What this study adds?

Similar to the other countries, congenital lungs malformations are also not commonly found in Thailand. The surgical outcomes are good with low complications. Moreover, our results showed the most patients usually had presenting symptoms from respiratory tract complications especially in neonatal period that need surgical intervention. Thus, our institute prefers to perform early operation. However, the clinical observation in asymptomatic patients can be done with closely monitor to prevent complications that may occur.

Potential conflicts of interest

None.

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ผลการรักษาโดยวิธีการผ่าตัดในผู้ป่วยโรคปอดผิดปกติแต่กำเนิด

ชนัญญา กรุณาสุเมตตา, ชูศักดิ์ คุปตานนท์, สมภพ พระธานี, วรวิทย์อินทนู, ชวลิต วงศ์พุทธะ

วัตถุประสงค์: *เพื่อศึกษาผลการรักษาโดยวิธีการผ่าตัดในผู้ป่วยที่เป็นโรคปอดผิดปกติแต่กำเนิด รวมถึงลักษณะและการดำเนินของโรคที่โรงพยาบาลศรีนครินทร์*

วัสดุและวิธีการ: *เป็นการศึกษาเวชระเบียนย้อนหลังของผู้ป่วยจำนวน 25 ราย ที่ได้รับการวินิจฉัยว่าเป็นโรคปอดผิดปกติแต่กำเนิดและได้รับการรักษาโดยการผ่าตัด ตั้งแต่ พ.ศ. 2544 ถึง พ.ศ. 2554*

ผลการศึกษา: *จากผู้ป่วยทั้งหมด 25 ราย ที่ได้รับการวินิจฉัยว่าเป็นโรคปอดผิดปกติแต่กำเนิดและได้รับการรักษาโดยการผ่าตัด พบว่าเป็นเพศชายจำนวน 16 ราย (64%) และเป็นเพศหญิง 9 ราย (36%) มีอายุที่ค่ามัธยฐาน 7 เดือน และน้ำหนักที่ค่ามัธยฐาน คือ 6 กิโลกรัม ผู้ป่วยได้รับการวินิจฉัยว่าเป็น CCAM 12 ราย (48%) pulmonary sequestration 7 ราย (28%) congenital lobar emphysema 4 ราย (16%) และ bronchogenic cyst 1 ราย (4%) โดยอาการแสดงที่พบมากที่สุดคืออาการติดเชื้อในระบบทางเดินหายใจซึ่งพบ 14 ราย (56%) หายใจลำบาก 7 ราย (28%) ฝีในปอด 1 ราย (4%) ไอเป็นเลือด 1 ราย (4%) ไม่มีอาการ 2 ราย (8%) ไม่มีผู้ป่วยที่เสียชีวิตใน 30 วันหลังการผ่าตัด ภาวะแทรกซ้อนหลังการผ่าตัดประกอบด้วย ปอดติดเชื้อ ปอดแฟบ ภาวะเลือดออกหลังการผ่าตัดและแผลติดเชื้อ การติดตามการรักษาหลังการผ่าตัด (ติดตามในระยะ 1-8 ปี) พบว่าผู้ป่วยทั้งหมดมีอาการปกติและไม่มีปัญหาเกี่ยวกับระบบทางเดินหายใจ*

สรุป: *การรักษาโรคปอดผิดปกติแต่กำเนิดโดยวิธีการผ่าตัดเป็นวิธีการรักษาที่ได้ผลดีและมีภาวะแทรกซ้อนน้อย*