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ORIGINAL ARTICLE

Thoracoscopic resection of congenital cystic lung lesions is associated with better post-operative outcomes

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Abstract
Introduction The incidence of congenital cystic lung lesions has been increasing in recent years due to better antenatal detection. With the introduction and maturation of thoracoscopy, the operative management for these lesions has seen advancement in the last decade. In this study, we aimed to compare the post-operative outcomes of patients who had thoracoscopic resection with those who underwent open resection.
Methods A retrospective review of all patients who underwent surgery for congenital cystic lung lesions between January 1996 and June 2012 in a tertiary referral center was conducted. Patients' demographics, operative procedures and post-operative outcomes were analyzed.
Results Sixty-seven patients were identified over the past 15 years. Thirty-nine patients had thoracoscopic resections and 28 had open resections. Thirteen patients in the thoracoscopic group required conversion. Both groups had similar demographics in terms of age, body weight and laterality of lesions. The mean operative time and blood loss in the two groups were comparable. Patients in the thoracoscopic group had significantly shorter duration of chest tube drainage (4.3 vs. 6.9 days, $p = 0.004$), shorter intensive care unit stay (2.5 vs. 5.9 days, $p = 0.003$) and shorter hospital stay (6.9 vs. 12.0 days, $p < 0.001$). Post-operative complication rate was similar between the two groups. Patients with body weight less than 5 kg showed a significantly higher conversion to open surgery as compared to those with body weight more than 5 kg (62.5 vs. 25.8 %, $p = 0.049$).
Conclusion Successful thoracoscopic resection for congenital cystic lung lesions results in better post-operative outcomes. However, this technique remains technically challenging in patients with body weight less than 5 kg.

Keywords Minimal invasive surgery · Thorax · Cystic lung · Infants · Body weight

Introduction

Major types of congenital cystic lung lesions include congenital cystic adenomatoid malformation (CCAM), bronchopulmonary sequestration, congenital lobar emphysema and bronchogenic cyst. These lesions can produce a wide variety of clinical presentation ranging from neonatal respiratory distress to remaining completely asymptomatic. With the increasing use of antenatal ultrasound screening, more and more lesions are picked up as purely incidental finding. Despite remaining asymptomatic at birth, these patients are prone to develop chest infection in childhood and carry potential risk of malignant transformation later in life [1]. Complete surgical resection is therefore the current standard of treatment. Most surgeons now advocate anatomical lobectomy, as non-anatomical wedge resection is associated with a higher incidence of residual lesion or recurrence [2].

Minimally invasive surgery is now the gold standard treatment in many surgical conditions. Thoracoscopic surgery in children has also come a long way since its first application in the 1970s [3]. With recent advances in endoscopic instruments and operative techniques, more technically demanding procedures such as thoracoscopic

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lobectomy are now safe and feasible [4–6]. The advantages of thoracoscopic procedures over open thoracotomy include less post-operative pain, early removal of chest drain, shorter hospital stay and better cosmetic outcome. In addition, thoracoscopic approach can reduce the incidence of musculoskeletal complications, such as scoliosis and chest wall deformities seen in conventional operation [7].

Till now only a few comparative studies have evaluated the post-operative outcomes between thoracoscopic and open thoracotomy approaches [8–13]. This study was undertaken to review our experience of thoracoscopic resection for congenital cystic lung lesions and to compare the outcomes with those who underwent open resection.

Patients and methods

The charts of all patients who underwent surgery for congenital cystic lung lesions at our center between January 1996 and June 2012 were retrospectively reviewed. No patient was excluded from the study. Pre-operative diagnosis was made either by routine antenatal ultrasound screening or after chest symptoms arise postnatally. All patients had chest radiography and computed tomography of thorax with contrast enhancement to confirm the diagnosis and location of the lesion before operation. The diagnosis of congenital cystic lung lesions was confirmed by histological examination of resected specimens.

Patients' demographics, peri-operative and post-operative outcomes were collected. Demographic data included sex, antenatal diagnosis, presence of pre-operative symptoms (respiratory distress or pneumonia), age at time of operation, body weight at time of operation, laterality of lesion, anatomical location of lesion, histopathology of lesion and follow-up time. Peri-operative outcomes included elective versus emergency operation, thoracoscopic versus open thoracotomy resection, the need for conversion to thoracotomy, operative blood loss, operative time and intra-operative complications. Post-operative outcomes included the duration of ventilator support, the duration of intensive care unit (ICU) stay, duration of chest tube drainage, post-operative complications and duration of hospitalization.

For thoracoscopic lobectomy, the patient was placed in lateral decubitus position. Single lung ventilation was achieved by standard endotracheal intubation together with the aid of endobronchial blocker or selective intubation of the contralateral bronchus. Pneumothorax was created by low flow (1 L/min) and low pressure (4 mmHg) carbon dioxide insufflation to enhance lung collapse and adequate exposure. Three 5 mm ports were placed, one for the 5 mm 30° telescope and two others as working channels. One additional 3 or 5 mm port may be required for retraction. Dissection was aided by the use of Ligasure (ValleyLab, Boulder, CO). Hemolok (Weck Closure Systems, Research Triangle Park, NC) were used to secure large vessels or bronchi. One of the port sites was enlarged to 2 cm to allow for removal of specimen. A chest drain was placed routinely after operation. For conventional open thoracotomy, the standard posterolateral approach over the fifth intercostal was employed. All patients were transferred to ICU post-operatively for observation overnight.

Statistical analysis of data was performed using SPSS (Version 17; SPSS, Chicago, IL). Continuous variables were analyzed using Student's t test. Ordinal variables such as follow-up time, duration of ventilator support, duration of ICU stay, duration of chest tube drainage and duration of hospitalization were analyzed using Mann–Whitney U test. Categorical variables were analyzed using Chi-square test. Data were presented as mean $\pm$ standard error of mean and range. $p < 0.05$ was considered statistically significant. Intention to treat approach was applied to data analysis. All those patients having thoracotomy converted from an initial thoracoscopic attempt were considered in the thoracoscopic group, such that a more conservative result would be generated.

Results

Sixty-seven patients underwent surgery for congenital cystic lung lesions at our unit between January 1996 and June 2012. Thoracoscopic resection was first introduced in 2002 and gradually became the approach of choice over the years (Fig. 1). Thirty-nine patients underwent thoracoscopic resection, while 28 patients underwent open thoracotomy. The two groups had comparable demographic data (Table 1). The mean age for the thoracoscopic resection group was 10.4 months (range 0.6–72 months) compared to 11.7 months (range 0.2–108 months) for thoracotomy group. The mean body weight at operation for thoracoscopic and thoracotomy groups were 8.1 kg (range 3.4–23.8 kg) and 7.0 kg (range 1.3–26.4 kg), respectively. Both the age and weight at operation showed no statistically significant difference between the groups. The sex ratio and laterality of lesions were similar. Four patients from the thoracoscopic group had respiratory symptoms before surgery (2 respiratory distress and 2 pneumonia), while there were 7 patients with symptoms (2 respiratory distress and 5 pneumonia) in the thoracotomy group. Majority of operations were performed electively in both groups (90 vs. 75 %, $p > 0.05$). CCAM was the most common pathology identified (47 of 67 patients).

There was no statistically significant difference in the operative time, intra-operative blood loss and duration of post-operative ventilatory support between the two groups. The mean operative time was 173 min (range 30–415 min)

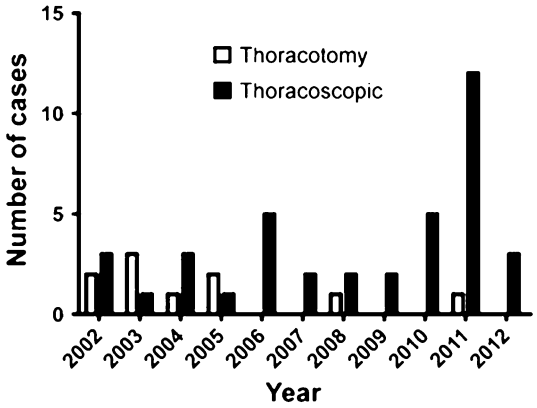

Fig. 1 Summary of the number of thoracoscopic resection and thoracotomy performed each year from 2002 to 2012. Data for 2012 is till 30th June

Table 1 Patients' demographics

	Thoracoscopic	Thoracotomy	*p*
Numbers	39	28	
Mean age at operation (months)	10.4 ± 2.3 (0.6–72)	11.7 ± 4.3 (0.2–108)	NS
Mean body weight at operation (kg)	8.1 ± 0.6 (3.4–23.8)	7.0 ± 1.1 (1.3–26.4)	NS
Sex: male (%)	19 (49 %)	23 (82 %)	NS
Laterality: left (%)	21 (54 %)	11 (39 %)	NS
Presence of pre-operative symptoms (%)			
Respiratory distress	2 (5 %)	2 (5 %)	NS
Pneumonia	2 (5 %)	5 (18 %)	NS
Elective surgery	35 (90 %)	21 (75 %)	NS
Histopathology (%)			
CCAM	25 (64 %)	22 (79 %)	
Extralobar sequestration	6 (15 %)	2 (7 %)	
Intralobar sequestration	7 (18 %)	1 (4 %)	
Congenital lobar emphysema	1 (3 %)	1 (4 %)	
Bronchogenic cyst	0	2 (7 %)	

NS Not significant

for thoracoscopic group and 172 min (range 85–320 min) for thoracotomy group. Blood loss and ventilatory support were 62.8 ml (range 0–400 ml) versus 67.1 ml (0–350 ml) and 0.49 days (range 0–2 days) versus 0.67 days (range 0–6 days), respectively.

The thoracoscopic group had a statistically significant shorter ICU stay [2.54 days (range 1–8 days) vs. 5.92 days (range 1–20 days), $p = 0.003$], duration of chest tube drainage [4.26 days (range 0–9 days) vs. 6.88 days (range 2–18 days), $p = 0.004$] and hospital stay [6.95 days (range 2–13 days) vs. 11.96 days (range 1–30 days), $p < 0.001$].

Six patients in each group experienced post-operative complications, including wound infection, surgical emphysema, pneumothorax, chylothorax and transient unilateral phrenic nerve palsy (Table 2). All the complications except one were treated conservatively. Chest drain re-insertion for the patient with pneumothorax after thoracoscopic resection was the only intervention required.

Thirteen (33 %) patients were converted to thoracotomy after initial thoracoscopic attempt. The main reasons for conversion included unstable intra-operative condition (desaturation or bleeding, 5 cases), poor visibility (4 cases), dense adhesions and difficult dissection (3 cases). One patient underwent conversion because the bronchial stump was too big for safe ligation thoracoscopically. On calculating the sliding average of 6 patients for the conversion rate, a clear decreasing trend was observed (Fig. 2). Subgroup analysis revealed that the incidence for conversion to thoracotomy was higher among patients less than 5 kg when compared with those above 5 kg (62.5 vs. 25.8 %, $p < 0.05$).

Discussion

The safety and feasibility of thoracoscopic resection of congenital cystic lung lesions have been documented in many published series [4, 5, 14]. Although these reported a number of advantages for the minimal invasive approach over conventional thoracotomy, few studies truly compared and evaluated the difference in post-operative outcomes.

Previous comparative studies produced conflicting results among some of the most important post-operative outcome measures, including operative time, duration of chest tube drainage, length of hospitalization and post-operative complications. Due to the limited number of patients and publications, there was no consensus on these issues. For instance, Rahman and Lakhoo [11] and Vu et al. [13] reported a longer operative time for thoracoscopic approach, while Bratu et al. [8], Bonnard et al. [12], Diamond et al. [9] and Tolg et al. [10] reported no difference. A meta-analysis recently published by Nasr and Bass [15] integrated data from these retrospective cohort and tried to produce a pooled estimate. Their study showed that the operative time was similar among the two approaches. Our study echoed the above findings, with a mean difference of only 1 min between the operative times of the two procedures. In fact, the operative time for thoracoscopic approach may still be over-estimated as the total time for initial attempted thoracoscopic resection and the subsequent open thoracotomy were included under the intention to treat study design. Similarly, Diamond et al. [9] and Rahman and Lakhoo et al. [11] did not show an advantage for thoracoscopic resection in terms of chest drain removal

Table 2 Operative outcomes

	Thoracoscopic	Thoracotomy	*p*
Mean operative time (min)	173 ± 13.9 (30–415)	172 ± 11.9 (85–320)	NS
Mean intra-operative blood loss (ml)	62.8 ± 14.5 (0–400)	67.1 ± 16.6 (0–350)	NS
Mean duration of ventilatory support (day)	0.49 ± 0.11 (0–2)	0.67 ± 0.26 (0–6)	NS
Mean ICU stay (day)	2.54 ± 0.30 (1–8)	5.92 ± 0.87 (1–20)	0.003
Mean duration of chest tube drainage (day)	4.26 ± 0.34 (0–9)	6.88 ± 0.87 (2–18)	0.004
Mean duration of hospitalization (day)	6.95 ± 0.42 (2–13)	11.96 ± 1.45 (1–30)	<0.001
Post-operative complication (%)	6 (15 %)	6 (21 %)	NS
Wound infection	2	2	
Surgical emphysema	2	1	
Pneumothorax	1	2	
Chylothorax	0	1	
Transient phrenic nerve palsy	1	0	

NS Not significant

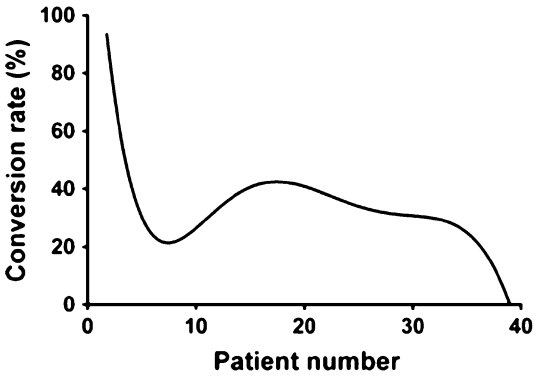

Fig. 2 Conversion rate for thoracoscopic lung resections during the study period (sliding average over 6 patients)

and hospitalization. In our series, chest drain removal and discharge from hospital were significantly earlier in patients who had thoracoscopic approach.

Significant shortening of post-operative ICU support provides more opportunities for other major operations. For our patients, chest tubes were removed only after the output became minimal and no pneumothorax was noted after clamping. This may explain why the average hospital stay after thoracoscopic resection in our series was slightly longer than those described in the literature. Cultural and institutional difference in terms of parental anxiety and exclusive government financial support may also be partly responsible.

Musculoskeletal complications including chest wall asymmetry, rib fusion, winged scapula and scoliosis are major long-term complications of thoracotomy and have been documented to occur in up to 30 % of patients in previously published series [7, 16]. Indeed, this genuine advantage of thoracoscopic approach over open thoracotomy was observed in our series as none of the patients in the thoracoscopic group developed the above-mentioned musculoskeletal complications. Significant improvement in cosmetic outcome also means much better patients' and parent's satisfaction.

The reason for conversion in our series was mainly due to intra-operative respiratory instability, difficulty in control of bleeding source, dense adhesions and difficult dissection due to fused fissures. Patients' safety should always be the surgeons' top priority, and there should be no hesitation in conversion for a smooth and safe operation. With advances in anesthetic technique and availability of new electrical devices, together with the growth of surgical experience, our team has now progressed through the learning curve (Fig. 2) and has achieved zero conversion rate in the latest year. Previously, Vu et al. suggested that pre-operative respiratory symptom was a risk factor for conversion due to its potential to create intra-thoracic adhesions. However, we failed to observe this association as all our patients who required conversion were asymptomatic. As mentioned previously, poor visualization and difficult mobilization of lung due to limited working space can be the major reasons for conversion. Indeed, in subgroup analysis, we demonstrated a lower conversion rate in patients more than 5 kg at the time of operation. This may serve as a guideline for future patient selection.

This study is by far one of the largest comparative studies among all the similar published series and we concluded that thoracoscopic approach is associated with better perioperative outcomes and similar post-operative complication rate when compared with thoracotomy. We recommend thoracoscopic approach for resection of congenital cystic lung lesions as the operation can be performed safely and effectively in specialized centers with good surgical expertise and anesthetic support. Thoracoscopic resection in patients with body weight less than 5 kg remains a technical challenge for surgeons.

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