Comparing surgical outcomes of complete thoracoscopic lobectomy for congenital cystic lung disease between neonatal and infantile patients

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ABSTRACT

Thoracoscopic lobectomy has recently become a widely used surgical treatment for congenital cystic lung disease, but significant issues can arise in some cases, such as a limited working space in neonates, a limited view in cases involving large cystic lesions. We reviewed the treatment outcomes of neonates that underwent complete thoracoscopic lobectomy or segmentectomy and evaluated the operative difficulties. From January 2008 to October 2015, 38 patients under the age of 1 year underwent complete thoracoscopic lobectomy or segmentectomy for cystic lung disease at our institution. We compared the intra- and postoperative data of the neonate group (N group) with those of the infant group (I group). Fourteen and 24 patients underwent thoracoscopic lobectomy or segmentectomy in the N group and I group, respectively. The operative time and amount of intraoperative blood loss did not differ significantly between the two groups (p=0.694 and p=0.878, respectively), but the duration of the postoperative hospitalization period was significantly longer (p<0.01) in the N group. The frequencies of postoperative complications did not differ significantly between the two groups. The operative time of thoracoscopic lobectomy was significantly longer in cases involving incomplete lobar fissures than in those involving normal lobar fissures. Surgical outcomes of complete thoracoscopic lobectomy for neonatal cases are almost equivalent compared with infantile cases, and thoracoscopic lobectomy takes longer in cases involving incomplete lobar fissures.

Key Words: thoracoscopic lobectomy, neonate, congenital lung disease, incomplete lobar fissure

INTRODUCTION

Minimally invasive surgery for congenital cystic lung disease in children has become more common in recent years. The frequency of prenatally diagnosed cases of these diseases is increasing, and many patients are indicated for thoracoscopic treatment. However, significant issues can arise in some cases, such as a limited working space in neonates, a limited view due to the presence of large cystic lesions, incomplete lobar fissures, and severe inflammatory adhesion after pneumonitis. There have been a few studies about the outcomes of thoracoscopic lobectomy...
PATIENTS AND METHODS

From January 2008 to December 2015, 38 patients under the age of 1 year underwent lung resection for congenital cystic lesions at our institution. The types of disease exhibited by the subjects included congenital pulmonary airway malformation (CPAM) (34 cases, including 3 cases that were complicated with pulmonary sequestration), intrapulmonary sequestration (3 cases), and bronchial atresia (1 case). The patients were divided into two groups; i.e., into patients that underwent complete thoracoscopic lobectomy or segmentectomy at <30 days of age (N group) and those that underwent such procedures at >30 days of age (I group). The patients were also divided into two other groups; i.e., into patients with incomplete lobar fissures and patients with normal lobar fissures. Incomplete lobar fissures were defined as a condition characterized by failing to recognize any lobar fissure. In addition, the cases with incomplete lobar fissure were divided into the N group and the I group. We retrospectively reviewed and compared various intraoperative and postoperative outcomes, including the operative time, amount of intraoperative blood loss, conversion rate, the duration of the postoperative hospitalization period, postoperative complications, and the frequency of postoperative pectus excavatum that required surgical treatment, among these groups. The Mann-Whitney U test and Fisher’s exact test were used to compare the clinical data between the two groups, and p-values of <0.05 were considered to be statistically significant.

Thoracoscopic technique

The patient was placed in the lateral position, and tracheal intubation was performed with a cuffed endotracheal tube to achieve bilateral lung ventilation. A 5-mm bladeless trocar was initially inserted under thoracoscopic observation with a 5-mm, 0° telescope to prevent lung injuries and the leakage of carbon dioxide around the port. The pleural cavity was initially inflated at a pressure of 8 mmHg and a flow rate of 0.5 L/min, and was then maintained at a pressure of 4–6 mmHg. Two or three additional ports were then inserted. The 5-mm, 30° telescope was inserted through the first port. The exact procedure varied in each case depending on the lobe that was resected. Any adhesion between the affected lung and chest wall was lysed. In cases involving incomplete fissures, the lung parenchyma was divided with laparoscopic coagulating shears or sealed with laparoscopic vessel ligation devices. The lobar vessels were dissected out and divided with laparoscopic vessel ligation devices and endoscopic clips. The bronchi were closed and divided with endoscopic clips. The specimens were removed using a disposable specimen pouch through a 12-mm port, which was produced via the enlargement of the initial 5-mm port incision. A chest tube was left in place in all cases.

RESULTS

The N group included 14 patients, whereas the I group included 24 patients (Table 1). The patients’ body weight at the time of surgery was significantly lower in the N group (p<0.01). The frequencies of prenatally diagnosed patients did not differ significantly between the two groups (14 [100%] vs. 21 [88%], p=0.283). In the N group, the lesions were located in the upper right
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Table 1  Surgical outcomes of complete thoracoscopic lobectomy for congenital cystic lung disease (neonates vs. infants)

|                          | Neonates (n=14) (age <30 days) | Infants (n=24) (age >30 days) | P-value |
|--------------------------|---------------------------------|--------------------------------|---------|
| Sex                      | Male:Female, 8:6                 | 9:15                           | 0.318   |
| Age at the time of surgery (days) | 25 (0–28)                     | 128 (31–362)                  | <0.01   |
| Birth weight (g)         | 3083±325                       | 2981±341                      | 0.545   |
| Weight at the time of surgery (g) | 3611±478                     | 6280±1543                     | <0.01   |
| Prenatal diagnosis (%)   | 14 (100%)                      | 21 (88%)                      | 0.283   |
| Gestational age at the time of the prenatal diagnosis (weeks) | 25.5 (20–34)               | 23 (20–32)                    | 0.164   |
| Maximum diameter of the cystic lesion (mm) | 41                            | 34                            | 0.495   |
| Maximum diameter of the cystic lesions (mm) / body weight (kg) | 10                            | 5.1                           | <0.01   |
| Preoperative infection   | 0 (0%)                          | 2 (8.3%)                      | 0.522   |
| Associated anomalies     | 2 (14.3%)                      | 2 (8.3%)                      | 0.616   |
| Follow-up period (months) | 51 (1.6–95)                    | 42 (1.2–83)                   | 0.449   |
| Operative time (min)     | 212 (84–441)                   | 177 (91–307)                  | 0.694   |
| Intraoperative blood loss (ml) | 5.0 (0–114)               | 5.5 (0–60)                    | 0.878   |
| Emergency operation (%)  | 3 (21%)                        | 0 (0%)                        | 0.043   |
| Conversion (%)           | 1 (7.1%)                       | 0 (0%)                        | 0.368   |
| Incomplete fissure (%)   | 5 (35.7%)                      | 7 (29.2)                      | 0.728   |
| Length of the postoperative hospitalization period (days) | 19.5 (7–88)          | 7 (4–41)                      | <0.01   |
| Major postoperative complications | 3 (21.4%)                    | 2 (8.3%)                      | 0.337   |
| Atelectasis              | 1                               | 1                              |         |
| Bronchial air leakage    | 2                               | 0                              |         |
| Middle lobe torsion      | 0                               | 1                              |         |
| Pectus excavatum         | 1 (7.1%)                       | 1 (4.2%)                      | 1.000   |

In 2 cases, the middle right lung in 2 cases, the lower right lung in 2 cases, the upper and middle right lung in 1 case, the upper left lung in 4 cases, and the lower left lung in 3 cases, whereas in the I group they were located in the upper right lung in 3 cases, the lower right lung in 11 cases, the upper and middle right lung in 1 case, the upper left lung in 2 cases, and the lower left lung in 7 cases. In the N group, thoracoscopic lobectomy, and segmentectomy was performed in 13, and 1 cases, respectively. In the I group, all patients underwent thoracoscopic lobectomy. Maximum diameter of the cystic lesion is similar between two group (41mm vs. 34mm, p=0.495), however maximum diameter of the cystic lesion / body weight ratio is much in the N group (10 vs. 5.1, p<0.01). The patients’ perioperative data and postoperative complications are shown in Table 1. The operative time and amount of intraoperative blood loss did not differ significantly between the two groups (212 vs. 177 min, p=0.694; 5.0 vs. 5.5 ml, p=0.878,
respectively). The frequency of emergency operations was significantly higher in the N group (3 [21%] vs. 0 cases [0%], p=0.043), and all three of these procedures were required because of a worsening respiratory status. Two cases had preoperative infection in the I group, one case had mild adhesion, and the other case had severe adhesion around the cystic lesions. In the I group, none of the patients suffered respiratory symptoms, and elective surgery was planned in each case. Only one patient (7.1%) in the N group underwent conversion to open lobectomy. The duration of the postoperative hospitalization period was significantly longer in the N group (19.5 vs. 7 days, p<0.01). The postoperative frequencies of major complications did not differ significantly between the two groups (21.4 % vs. 8.0 %, p=0.337). Major complications occurred in 3 cases in the N group (atelectasis in 1 case and bronchial air leakage in 2 cases) and 2 cases in the I group (atelectasis in 1 case and middle lobe torsion in 1 case). Two patients in the N group underwent thoracoscopic reoperation due to bronchial air leakage. The frequency of postoperative pectus excavatum that required surgical treatment did not differ between the two groups.

A comparison of the treatment outcomes of the patients with and without incomplete lobar fissure including neonatal and infantile cases is shown in Table 2. The operative time was significantly longer in the cases involving incomplete fissures. However, amount of intraoperative blood loss, the duration of the postoperative hospitalization period, and the postoperative frequencies of major complications did not differ between the two groups (7 vs. 5 ml, p=0.319;

| Table 2 | Surgical outcomes of complete thoracoscopic lobectomy for congenital cystic lung disease (incomplete fissure vs. non-incomplete fissure, age<1 year) |
|---------|--------------------------------------------------------------------------------|
| Patients with incomplete fissures (n=12) | Patients without incomplete fissures (n=26) | P-value |
| **Sex** | | | |
| Male:Female | 4:8 | 13:13 | 0.486 |
| **Body weight at the time of surgery (g)** | 4996±1875 | 5436±1793 | 0.451 |
| **Age at the time of surgery (days)** | 49 (49–362) | 77 (0–362) | 0.593 |
| **Prenatal diagnosis (cases)** | 11 (99%) | 24 (92%) | 1.000 |
| **Preoperative infection** | 1 (8.3%) | 1 (3.8%) | 0.538 |
| **Associated anomalies** | 0 (0%) | 4 (15%) | 0.287 |
| **Follow-up period (months)** | 38.3 (1.2–73) | 42 (1.6–95) | 0.414 |
| **Operative time (min)** | 267 (88–312) | 158 (84–347) | **0.014** |
| **Intraoperative blood loss (ml)** | 7 (1–76) | 5 (0–114) | 0.319 |
| **Emergency operation (%)** | 1 (8.3%) | 2 (7.7%) | 1.000 |
| **Conversion (%)** | 0 (0%) | 1 (3.8%) | 1.000 |
| **Length of the postoperative hospitalization period (days)** | 8 (6–55) | 10 (4–88) | 0.559 |
| **Postoperative complications** | 1 (8.3%) | 4 (15%) | 1.000 |
| Atelectasis | 1 | 1 |
| Bronchial air leakag | 0 | 2 |
| Middle lobe torsion | 0 | 1 |
| Pectus excavatum | 0 (0%) | 2 (7.7%) | 1.000 |
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8 vs. 10 days, p=0.559; 1 vs. 4 cases, p=1.000, respectively).

A comparison of the treatment outcomes of the patients with incomplete lobar fissure between the N and the I group is shown in Table 3. The operative time was relatively equivalent in the neonatal cases involving incomplete fissures (224 min vs. 274, p=0.811). The intraoperative blood loss, the duration of the postoperative hospitalization period, and the postoperative frequencies of major complications were also equivalent between the two groups (10 vs. 5 ml, p=0.567; 12 vs. 7 days, p=0.06; 1 vs. 0 case, p=0.417, respectively).

### DISCUSSION

Pediatric cystic lung diseases encompass a spectrum of congenital and acquired disorders, with the most common cause of congenital lung cysts being CPAM. Ten-40% of patients with CPAM will develop significant pulmonary infections, and cases of malignant lesions associated with CPAM have also been reported. Some authors consider that the complete excision of congenital lung lesions should be the standard treatment for pediatric cystic lung disease, although other authors are much more cautious and recommend intervention only after the development of symptoms or complications, even if the patient is diagnosed during the prenatal period. A meta-analysis of the postoperative management of congenital cystic lung lesions showed that elective surgery is associated with better outcomes than emergency surgery although the risk of

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### Table 3 Surgical outcomes of complete thoracoscopic lobectomy for congenital cystic lung disease with incomplete lobar fissure (neonates vs. infants)

|                              | Neonates with incomplete lobar fissure (N=5) | Infants with incomplete lobar fissure (N=7) | P value |
|------------------------------|---------------------------------------------|---------------------------------------------|---------|
| Sex                          | Male:Female 2:3                              | 2:5                                         | 1.000   |
| Age at the time of surgery (days) | 25 (1–28)                                  | 77 (60–362)                              | **<0.01** |
| Weight at time of surgery (g)    | 3557±357                                    | 6025±1845                                  | **<0.01** |
| Prenatal diagnosis (%)         | 5 (100%)                                    | 6 (86%)                                    | 1.000   |
| Preoperative infection         | 0 (0%)                                       | 1 (14%)                                    | 1.000   |
| Associated anomalies           | 0 (0%)                                       | 0 (0%)                                     | 1.000   |
| Follow-up period (months)      | 42 (1.8–73)                                 | 35 (1.2–50)                               | 0.730   |
| Operative time (min)           | 224 (206–441)                               | 274 (88–317)                              | 0.811   |
| Intraoperative blood loss (ml) | 10 (2–76)                                    | 5 (1–60)                                   | 0.567   |
| Emergency operation (%)        | 1 (20%)                                      | 0 (0%)                                     | 0.417   |
| Conversion (%)                 | 0 (0%)                                       | 0 (0%)                                     | 1.000   |
| Length of the postoperative hospitalization period (days) | 12 (8–55)                                  | 7 (6–10)                                  | 0.06    |
| Major postoperative complications | 1 (20%)                                    | 0 (0%)                                     | 0.417   |
| Atelectasis                   | 1 (20%)                                      | 0 (0%)                                     | 0.417   |
| Pectus excavatum              | 0 (0%)                                       | 0 (0%)                                     | 1.000   |
symptoms occurring in previously asymptomatic cases is small.\textsuperscript{15)}

Thoracoscopic lung lobectomy for congenital cystic lung lesions is a well-described technique that represents an alternative to open lobectomy.\textsuperscript{1, 3, 4, 7, 16-18)} The feasibility of thoracoscopic lobectomy in small children has been established by numerous groups, culminating in recent meta-analyses demonstrating shorter hospital lengths of stay and comparable operative times, and feasibility alternative to open resection.\textsuperscript{17, 19)} Kulaylat also reported that thoracoscopic resection was associated with decreased postoperative complications and length of stay.\textsuperscript{20)} Many authors agree on the benefit of thoracoscopic procedures, e.g., they are associated with less pain; shorter hospital stays; better cosmetic results; and a decreased midterm morbidity rate, including lower frequencies of chest wall deformities, musculoskeletal deformities, and scoliosis.\textsuperscript{21)} However, significant issues can arise during such procedures in some cases, such as a small working space in neonates, a limited view due to large cystic lesions, incomplete lobar fissures, and severe inflammatory adhesion after pneumonia.\textsuperscript{7)} In infants, the small space within the thoracic cavity can result in technical difficulties during thoracoscopic operations. Kunisaki \textit{et al.} also reported that in infants and younger children thoracoscopic procedures were associated with longer operative times than thoracotomy.\textsuperscript{8, 17)} These findings support the notion that a definite learning curve exists for pediatric thoracoscopic lobectomy, as higher-volume thoracoscopic surgeons perform such procedures faster than lower-volume surgeons.\textsuperscript{17)} Laje said that thoracoscopic lobectomy was not a procedure that can be quickly mastered, required a unique skill set relative to most other advanced MIS procedure, and should be performed in institutions with adequate volume. In this study, operative time was similar between in earlier cases and in later cases (213 vs. 215min), even if in neonatal cases. In addition, we detected a higher complications rate in patients that suffered chest infections than in patients that were diagnosed prenatally.\textsuperscript{7)}

Few studies have compared the outcomes of thoracoscopic lobectomy for congenital cystic diseases between patients that underwent the procedure at <30 days of age and those that underwent the procedure at >30 days of age. There have also been few comparative studies of the results of thoracoscopic lobectomy involving patients between with and without incomplete lobar fissures, and between neonatal and infantile cases with incomplete fissures. In this study, neither the operative time nor amount of intraoperative blood loss differed significantly between the N group and I group. There is one conversion case in the N group, it was similar in comparison with Laje’s study (12%).\textsuperscript{8)} The length of the postoperative hospitalization period was significantly longer in the N group than in the I group. In neonatal patients who require emergency surgery due to a worsening of respiratory status, it can take a while for the patient’s respiratory status to improve. Neonates with other anomalies also require treatment. The postoperative frequency of major complications did not differ significantly between the two groups (3 [21.4%] vs. 2 cases [8.3%]). It was equivalent in comparison with Lajes’s study for infants (9%). However, reoperation was required in 2 cases of bronchial air leakage in the N group. The neonatal bronchi are so fragile that clinicians must take great care when identifying and ligating them.

The lack of space in the infant thoracic cavity can cause technical difficulties during thoracoscopic surgery. Actually, the size of the cystic lesions for body weight was much in neonatal cases. Large space-occupying lesions that are not decompressed by ventilation can be successfully decompressed using laparoscopic coagulating shears. In our experience, it is possible to obtain sufficient working space and a large enough operative field even in neonatal cases. Another challenge is controlling the vascular supply to the affected lobe.\textsuperscript{21)} This can be achieved using tiny endoscopic clips and laparoscopic vessel ligating devices.

This study for total cases found that it took longer than usual to dissect and divide incomplete lobar fissures during thoracoscopic lobectomy. However, this was not related to intraoperative bleeding or postoperative complications. We were not able to preoperatively determine whether the
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patients had incomplete lobar fissures using high-resolution computed tomography, so clinicians must keep the possibility that such anomalies might be present in mind during thoracoscopic lobectomy. Surgical outcomes in the N group with incomplete fissure was also equivalent compared with the I group.

In conclusion, our study showed that surgical outcomes of complete thoracoscopic lobectomy for congenital cystic lung disease in neonatal cases is almost equivalent compared with infantile cases, and that lobectomy takes longer in cases involving incomplete lobar fissures. In the latter patients, the follow-up period should be extended in order to detect late morbidities.

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