ORIGINAL PAPER

Nagoya J. Med. Sci. **78**. 447 ~ 454, 2016
doi:10.18999/nagjms.78.4.447

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

Takahisa Tainaka, Hiroo Uchida, Yujiro Tanaka, Chiyoe Shirota, Kazuki Yokota, Naruhiko Murase, Kazuo Oshima, Ryo Shirotsuki, Kosuke Chiba and Akinari Hinoki

Department of Pediatric Surgery, Nagoya University Graduate School of Medicine, Nagoya, Japan

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.[1-5] The frequency of prenatally diagnosed cases of these diseases is increasing,[6] 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.[4,7] There have been a few studies about the outcomes of thoracoscopic lobectomy

Received: June 23, 2016; accepted: August 26, 2016

Corresponding author: Hiroo Uchida, MD, PhD

Department of Pediatric Surgery, Nagoya University Graduate School of Medicine, 65 Tsurumai-cho, Showa-ku, Nagoya 466-8550, Japan

Phone: +81-52-744-2959, Fax: +81-52-744-2980, E-mail: hiro2013@med.nagoya-u.ac.jp

for children aged <1 year old.[8,9] The aim of this study was to evaluate the operative difficulties experienced in neonatal cases involving a small pleural cavity, and involving incomplete lobar fissures of congenital cystic lung disease.

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

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

lung 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<1year)

	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

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 equiavalent 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).

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

DISCUSSION

Pediatric cystic lung diseases encompass a spectrum of congenital and acquired disorders, with the most common cause of congenital lung cysts being CPAM.[10] Ten-40% of patients with CPAM will develop significant pulmonary infections,[11,12] and cases of malignant lesions associated with CPAM have also been reported.[13,14] 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.[11,14] 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

symptoms occurring in previously asymptomatic cases is small.[15]

Thoracoscopic lung lobectomy for congenital cystic lung lesions is a well-described technique that represents an alternative to open lobectomy.[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.[17, 19] Kulaylat also reported that thoracoscopic resection was associated with decreased postoperative complications and length of stay.[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.[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.[7] In infants, the small space within the thoracic cavity can result in technical difficulties during thoracoscopic operations. Kunisaki *et al.* also reported that in infants and younger children thoracoscopic procedures were associated with longer operative times than thoracotomy.[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.[17] Laje said that thoracosopic 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.[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%).[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.[2] 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

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.

ACKNOWLEDGMENT

The authors declare that they have no conflicts of interest.

REFERENCES

1) Albanese CT, Sydorak RM, Tsao K, Lee H. Thoracoscopic lobectomy for prenatally diagnosed lung lesions. *J Pediatr Surg*, 2003; 38: 553–555.
2) Cano I, Anton-Pacheco JL, Garcia A, Rothenberg S. Video-assisted thoracoscopic lobectomy in infants. *Eur J Cardiothorac Surg*, 2006; 29: 997–1000.
3) Diamond IR, Herrera P, Langer JC, Kim PC. Thoracoscopic versus open resection of congenital lung lesions: a case-matched study. *J Pediatr Surg*, 2007; 42: 1057–1061.
4) Rothenberg SS. First decade's experience with thoracoscopic lobectomy in infants and children. *J Pediatr Surg*, 2008; 43: 40–44.
5) Vu LT, Farmer DL, Nobuhara KK, Miniati D, Lee H. Thoracoscopic versus open resection for congenital cystic adenomatoid malformations of the lung. *J Pediatr Surg*, 2008; 43: 35–39.
6) Sauvat F, Michel JL, Benachi A, Emond S, Revillon Y. Management of asymptomatic neonatal cystic adenomatoid malformations. *J Pediatr Surg*, 2003; 38: 548–552.
7) Kaneko K, Ono Y, Tainaka T, Sumida W, Kawai Y, Ando H. Thoracoscopic lobectomy for congenital cystic lung diseases in neonates and small infants. *Pediatr Surg Int.* 2010; 26: 361–365.
8) Laje P, Pearson EG, Simpao AF, Rehman MA, Sinclair T, Hedrick HL, *et al.* The first 100 infant thoracoscopic lobectomies: Observations through the learning curve and comparison to open lobectomy. *J Pediatr Surg*, 2015; 50: 1811–1816.
9) Rothenberg SS, Kuenzler KA, Middlesworth W, Kay S, Yoder S, Shipman K, *et al.* Thoracoscopic lobectomy in infants less than 10 kg with prenatally diagnosed cystic lung disease. *J Laparoendosc Adv Surg Tech A*, 2011; 21: 181–184.
10) Stocker JT. Cystic lung disease in infants and children. *Fetal Pediatr Pathol.* 2009; 28: 155–184.
11) Aziz D, Langer JC, Tuuha SE, Ryan G, Ein SH, Kim PC. Perinatally diagnosed asymptomatic congenital cystic adenomatoid malformation: to resect or not? *J Pediatr Surg*, 2004; 39: 329–334.
12) Wong A, Vieten D, Singh S, Harvey JG, Holland AJ. Long-term outcome of asymptomatic patients with congenital cystic adenomatoid malformation. *Pediatr Surg Int*, 2009; 25: 479–485.
13) Ozcan C, Celik A, Ural Z, Veral A, Kandiloglu G, Balik E. Primary pulmonary rhabdomyosarcoma arising within cystic adenomatoid malformation: a case report and review of the literature. *J Pediatr Surg*, 2001; 36: 1062–1065.
14) Hsieh CC, Chao AS, Chang YL, Kuo DM, Hsieh TT, Hung HT. Outcome of congenital cystic adenomatoid malformation of the lung after antenatal diagnosis. *Int J Gynaecol Obstet*, 2005; 89: 99–102.
15) Stanton M, Njere I, Ade-Ajayi N, Patel S, Davenport M. Systematic review and meta-analysis of the postnatal management of congenital cystic lung lesions. *J Pediatr Surg*, 2009; 44: 1027–1033.
16) Tanaka Y, Uchida H, Kawashima H, Sato K, Takazawa S, Masuko T, *et al.* Complete thoracoscopic versus video-assisted thoracoscopic resection of congenital lung lesions. *J Laparoendosc Adv Surg Tech A*, 2013; 23: 719–722.
17) Kunisaki SM, Powelson IA, Haydar B, Bowshier BC, Jarboe MD, Mychaliska GB, *et al.* Thoracoscopic vs open lobectomy in infants and young children with congenital lung malformations. *J Am Coll Surg*, 2014; 218: 261–270.

18) Rothenberg SS, Shipman K, Kay S, Kadenhe-Chiweshe A, Thirumoorthi A, Garcia A, *et al.* Thoracoscopic segmentectomy for congenital and acquired pulmonary disease: a case for lung-sparing surgery. *J Laparoendosc Adv Surg Tech A*, 2014; 24: 50–54.
19) Nasr A, Bass J. Thoracoscopic vs open resection of congenital lung lesions: a meta-analysis. *J Pediatr Surg.* 2012; 47: 857–861.
20) Kulaylat AN, Engbrecht BW, Hollenbeak CS, Safford SD, Cilley RE, Dillon PW. Comparing 30-day outcomes between thoracoscopic and open approaches for resection of pediatric congenital lung malformations: Evidence from NSQIP. *J Pediatr Surg.* 2015; 50: 1716–1721.
21) Lawal TA, Gosemann JH, Kuebler JF, Gluer S, Ure BM. Thoracoscopy versus thoracotomy improves midterm musculoskeletal status and cosmesis in infants and children. *Ann Thorac Surg*, 2009; 87: 224–228.