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Uniportal Thoracoscopic Lobectomy for Congenital Lung Malformations in Children

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Abstract

Introduction: Single-incision paediatric endosurgery is gaining popularity, especially for abdominal operations. However, few reports in the literature support the feasibility of uniportal thoracoscopic surgery for congenital lung malformations in paediatric patients. This paper presented the initial experience with uniportal thoracoscopic surgery and compared the surgical outcomes with multiport Video-Assisted Thoracoscopic Surgery (VATS) and traditional thoracotomy.

Materials and Methods: From January 2009 to January 2018, a retrospective review of all patients (less than 18 years old) who underwent surgery for congenital lung malformations at our institution was performed. General data, operative data, anaesthesia parameters and surgical outcomes were collected. Results were analysed with one-way ANOVA and chi-square tests for continu-ous and categorical data.

Results: Twenty-one patients with congenital lung malformations underwent surgery at our institution. The number of patients enrolled in thoracotomy, multiportal VATS and uniportal VATS groups was 8, 6 and 7, respectively. No difference in age, body weight, post-operative stay and drainage period was observed among the groups. A considerably high conversion rate (3/6, 50%) was observed in the multiportal VATS group. After the introduction of endobronchial blocker for one lung ventilation, all multi-portal and uniportal VATS procedures could be performed safely without residual lesion or mortality.

Conclusions: VATS is feasible in the paediatric population, and it needs experienced surgical skills and unique anaesthetic requirements. For small infants with congenital lung malformations, uniportal VATS with endobronchial blocker is preferred.

Keywords: Congenital Pulmonary Malformations; Endobronchial Blocker; Uniportal Thoracoscopy

Introduction

The standard treatment for congenital lung malformation is anatomical resection [1,2]. Since the first case in 1993 and the first large series of Video-Assisted Thoracoscopic Surgery (VATS) lobectomy reported by Dr. Rothenburg in 2000 [3], minimal invasive surgeries for paediatric congenital lung lesions have become increasingly popular worldwide [4-6]. Single-incision, or uniportal VATS lobectomy in adults was first performed by Dr. Gonzalez in 2010 [7]. For children, the first uniportal VATS lobectomy was reported by Dr. Aragón in 2013 [8] about middle lobectomy for pulmonary aspergilloma. Few reports about uniportal VATS lobectomy or segmentectomy in children are found in the literature [9-13]. Modification to decrease collision between camera and instruments within the small intercostal space (ICS) in children has been proposed [14]. However, the reported cases involved children older than 1 year and who mostly had diseases rather than congenital lung malformations.

Thoracoscopy provides small incision and less stretching of ribs and intercostal muscles and may prevent late onset shoulder girdle weakness, loss of breast volume, chest wall deformity and spine deformity after thoracotomy in the future [15-17]. In multiportal VATS lobectomy, small working space and problems of angulations post more difficulties in infants and young children than in adolescents and adults [18]. Meanwhile, to remove specimen after multiportal VATS lobectomy, an elongated incision is often needed, and the elongated wounds are often sufficiently large for the passage of multiple instruments and direct management of vessels or incomplete fissures [12,19,20]. Therefore, uniportal VATS may have the potential to reduce incisions without compromising the accessibility of minimal invasive surgery of congenital lung anomalies. Herein, the preliminary outcomes of uniportal VATS procedures for congenital lung malformations were analyzed and compared with open thoracotomy and multiportal VATS.

Materials and Methods

From January 2009 to January 2018, total 21 patients underwent anatomical resection for congenital lung malformations: eight patients for open thoracotomy, six patients for multiportal VATS and seven for uniportal VATS. Most of the patients (14/21, 66.7%) had prenatal diagnosis or suspicion according to the ultrasound results. All patients except one preterm neonate with respiratory distress underwent pre-operative Computerised Tomography (CT) scan to confirm the diagnosis and surgical planning. Medical records, including operative age, body weight, diagnosis, operative procedure, operative time, anaesthesia parameters, one-lung ventilation techniques, post-operative recovery and complications, were analysed and compared (Table 1).

Group	Thoracotomy	Multiportal VATS	Uniportal VATS	P value
Patient number	8	6	7	
Male gender	7 (87.5%)	3 (50%)	3 (42.9%)	.160
Age (month)	33.9 (1 d-13 y)	14.4 (4 d-6 y)	31.1 (2 m-15 y)	.797
Body weight (kg)	10.7 (2.5-40)	7.8 (2.8-21)	12.7 (5.1-46)	.773
Comorbidity patient number	2* (25%)	1** (16.7%)	0	.599
Number of patients with symptoms	8 (100%)	2 (33.3%)	1 (14.3%)	.001
Prenatal diagnosis	4 (50%)	4 (66.7%)	6 (85.7%)	.376
Pre-op. haemoglobin (g/dL)	13.7±3.44	12.1±2.31	12.9±2.83	.626
Pre-op. platelet count ($10^3/\mu L$)	379.1±131.18	420.5±168.18	471.1±154.40	.508
Data expression: mean±standard deviation, mean (range) or number (%); * one prematurity, one cerebral palsy; ** prematurity; m: month old; d: day old; y: year old; pre-op.: pre-operative				

Table 1: General data of the patients in the three groups.

Shows the patients' characteristics in the three groups. Lung isolation is not required for open thoracotomy lobectomy at our institute. To achieve one-lung ventilation in VATS operation, three techniques were used in the study period: double-lumen endotracheal intubation, intrathoracic CO_2 insufflation (pressure 4 mmHg and flow rate 0.5-1 L/min) and endobronchial blocker.

Open thoracotomy was performed via 4_{th} or 5_{th} ICS with muscle-sparing posterolateral technique. Multiportal VATS was conducted via 4_{th}, 6_{th} and 8_{th} ICS (or 5_{th} and 7_{th} ICS) with or without trocar insertion. The specimen was placed into a sterilised plastic bag and removed via an elongated wound. The incision of uniportal VATS was created through 4_{th} or 5_{th} ICS in 2-3 cm length by using 5 mm or 3 mm laparoscopic instruments. Vessels were ligated with silk sutures. For bronchus resection and incomplete fissure division, staplers were used. The specimen could be directly removed without stretching wound or spreading ribs. Chest tube was routinely placed regardless of different approaches. The patients of VATS lobectomy all were transferred to the Intensive Care Unit (ICU) for postoperative care. In statistical analysis, one-way ANOVA test for continuous data and Fisher exact probability test for categorical data were used. All subjects enrolled in this study were approved by the Institutional Committee on Human Research of Taichung Veterans General Hospital, Taichung, Taiwan, in accordance with the guidelines of the Declaration of Helsinki and the International Conference on Harmonization for Good Clinical Practice (IRB TCVGH No. CE17254A).

Results

Nearly 70% of the patients had prenatal diagnosis based on regular ultrasound examinations; thus, they were born with stand-by neonatologists for possible respiratory distress. Three patients (3/21, 14.3%, not shown in the table) had respiratory conditions requiring ventilator support immediately after birth and received surgery in the neonatal period. One asymptomatic patient underwent surgery at the age of 4 days with a huge cystic lung lesion. Except for one patient with respiratory failure immediately after birth, all other patients had CT scan for to confirm the diagnosis and surgical planning. Most prenatally diagnosed patients underwent elective surgery at the age of 2 to 9 months, unless they became symptomatic earlier. Majority of the patients whose diagnoses were first made after birth were symptomatic (6/7, 85.7% versus patients with prenatal diagnosis and postnatal symptoms, 6/14, 42.9%), and the surgeries were performed soon after diagnoses. On the basis of different surgical approaches, pre-operative symptomatic rates differed with significance (p=.001). All patients in the thoracotomy group had pre-operative symptoms, such as cough, respiratory distress, tachypnea or infection-related illness. In the VATS groups, symptomatic rates were lower (multiportal/ uniportal, 33.3%/14.3%).

(Figure 1) shows the incisions for uniportal VATS. The distribution of involved lobes and pathological diagnoses did not differ among three groups (Table 2). Lower lobes were most frequently involved (17/21, 81.0%). Most cases involved congenital pulmonary airway malformations (CPAM) by the pathological report (11/21, 52.4%), which are similar to reported articles [3,21].

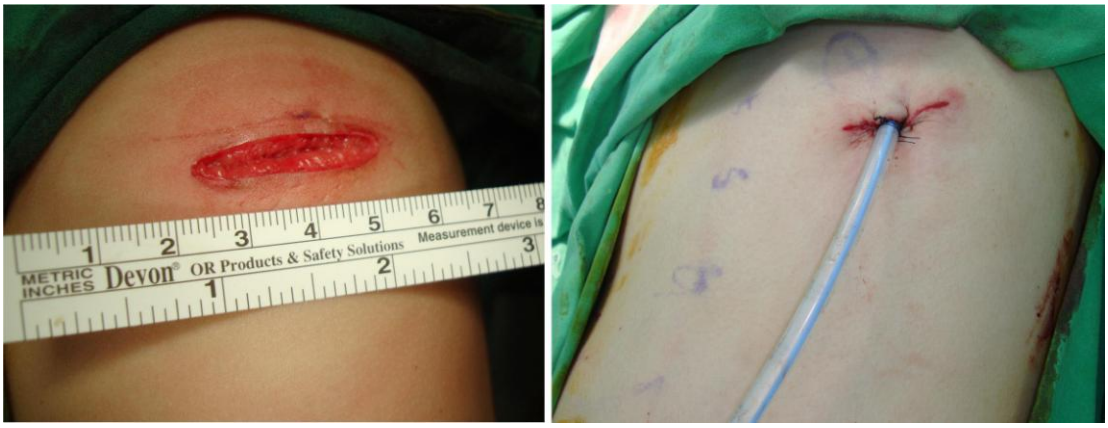

Figure 1: Incisions of uniportal VATS; left: 8-month-old, 7.9 kg intralobar pulmonary sequestration, LLL lobectomy; right: 15-year-old, 46 kg CPAM, left apicoposterior segmentectomy.

Group	Thoracotomy	Multiportal VATS	Uniportal VATS	P value
Involved lobes				
RLL	1	2	3	.491
LUL	1	1	1	1.000
LLL	4	2	3	.867
RUL + RML		1		.286
RML + RLL	2			.305
Resection methods				
Segmentectomy	2	1	2	1.000
Lobectomy	6	5	5	1.000
Pathological diagnosis				
Type-1 CPAM	1		2	.600
Type-2 CPAM	1	1	3	.443
CPAM, uncertain type	2	1		.600
CLO	2	1		.600
Bronchogenic cyst			1	.619

Intrapulmonary sequestration		1	1	.505
CPAM+sequestration	1	1		.733
Pneumatocele		1		.286
Pseudocyst	1			1.000

RUL: Right Upper Lobe; RML: Right Middle Lobe; RLL: Right Lower Lobe; LUL: Left Upper Lobe; LLL: Left Lower Lobe; CPAM: Congenital Pulmonary Airway Malformation, classification by Stocker in 2002 (Ref. 24), uncertain type due to destruction of distinct structure; CLO: Congenital Lobar Overinflation

Table 2: Patient numbers by involved lobes, anatomical resections and pathological diagnoses.

No conversion in uniportal VATS group was observed; however, three patients in multiportal VATS group eventually required thoracotomy for bleeding control, poor anaesthesia compliance (oxygen desaturation) and lack of working space for staplers. The average operative time was compatible in groups; however, significantly longer one-lung ventilation period was observed in uniportal VATS group than multiportal VATS (199.2 min versus 108.3 min, p=.019). The result was undoubtedly due to the early conversion in the multiportal VATS group (Table 3).

Group	Thoracotomy	Multiportal VATS	Uniportal VATS	P value
Conversion	NA	3 (50%)	0	.070
Operation duration (min)	194.4±87.11	238.3±88.24	270.0±46.46	.185
Anaesthesia duration (min)	220.0±91.07	295.8±114.95	328.6±58.72	.081
One-lung duration (min)	NA	108.3±53.26	*199.2±58.86	**.019**
Blood loss (cc/kg)	20.9 (0-79.0)	40.7 (0-38.1)	6.16 (0-12.7)	.066
Blood transfusion (cc/kg)	15.0 (0-52.6)	7.72 (0-30)	4.29 (0-12.7)	.366
Chest drainage period (day)	5.8±1.98	6.5±3.94	5.7±3.15	.872
ICU stay (day)	6.1±4.61	8.3±11.08**	5.1±5.43***	.726
Hospital stay (day)	12.3±8.76	14.2±11.23	7.9±4.88	.403
Analgesic shots (time)	3.4±2.62	1.3±1.21	3.7±1.70	.100
Follow-up period (month)	54.5 (4-88)	7.5 (1-17)	19.4 (2-44)	**.003**

Data expression: mean±standard deviation, mean (range) or number (%); N/A: not applicable; ICU: intensive care unit; *one patient of uniportal VATS group without one-lung ventilation; **one patient had prolonged intubation at ICU for 27 days; another preterm neonate had 17 days of ICU stay; ***one patient had unplanned extubation and 16 days of ICU stay

Table 3: Surgical parameters and post-operative recovery of groups.

Shows the surgical parameters and post-operative recovery. Blood loss and transfusion amounts were not significantly different in groups. However, the difference of blood loss in multiportal and uniportal VATS groups was observed (40.7 cc/kg versus 6.16 cc/ kg, p=.001). Chest drainage period was not influenced by surgical methods. Post-operative ICU and hospital stay was longer in multiportal VATS group without significance, less analgesic shots were given in the multiportal VATS group (1.3 shot versus 3.4 and 3.7 shots) and shorter follow-up period in the multiportal VATS group was also found. There is no lesion recurrence or mortality in all groups. In multiportal VATS group, most complications were related to conversion, as previously mentioned. Two major complications, delayed pneumothorax and tracheal stenosis, were recorded in the uniportal VATS group. Delayed pneumothorax was found 3 months after uniportal VATS left apicoposterior segmentectomy for type-1 CPAM in a 15-year-old female patient. Closed chest drainage with intrapleural chemical pleurodesis with OK-432 (5 KE Picibanil, Chugai Pharmaceutical Co., Ltd., Tokyo, Japan) was performed, and the patient had been symptom free for 4 years. Post-operative tracheal stenosis occurred in an 8-month-old male patient who initially had uniportal VATS LLL lobectomy for type-1 CPAM. Accidental extubation was encountered in ICU, and re-intubation was performed urgently due to respiratory distress. Three months later, the patient developed stridor, and bronchoscopy found shown in (Figure 2) subglottic stenosis.

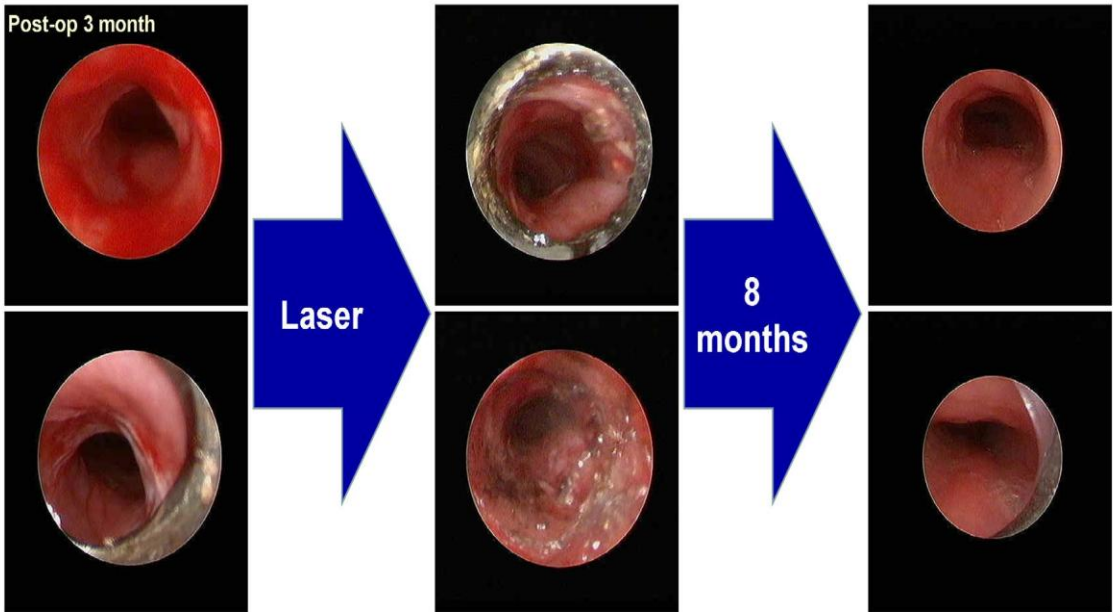

Figure 2: Post-operative tracheal stenosis: 8-month-old, 9 kg, uniportal VATS LLL lobectomy for type-1 CPAM.

The patient received laser ablation and had been asymptomatic for 2 years. One-lung ventilation methods differed in each group. (Table 4) lists the anaesthesia parameters.

Group	Thoracotomy	Multiportal VATS	Uniportal VATS	P value
One-lung ventilation methods				
Double lumen endotracheal tube	N/A		1 (14.3%)	1.000
CO_2 inflation	N/A	5 (83.3%)	1 (14.3%)	.080
Endobronchial blockers	N/A	1 (16.7%)	4 (57.1%)	.242
Anaesthesia parameters				
Lowest tidal volume (mL/kg)	7.7±1.22	7.9±1.73	7.1±1.24	.576
Lowest sPO_2 (%)	89.0±16.41	88.8±14.72	98.0±3.00	.341
Lowest mean arterial blood pressure (mmHg)	43.1±12.36	47.3±10.44	45.6±9.16	.775
Lowest body temperature (°C)	35.4±0.78	35.3±0.96	35.5±0.71	.912
Highest $etCO_2$ (mmHg)	43.9±7.80	47.3±5.92	41.6±3.05	.243
Highest airway pressure(cmH_2O)	24.4±4.47	24.2±7.29	27.4±3.78	.461
Lowest arterial blood pH	7.33±0.068	7.28±0.060	7.33±0.083	.516
Data expression: mean±standard deviation or number (%); N/A: not applicable				

Table 4: Patient numbers of one-lung ventilation methods and anaesthesia parameters.

Transient low peripheral oxygen saturation (sPO2) and high end-tidal carbon dioxide (etCO2) were noted in multiportal VATS patients with insignificant difference (Figure 3).

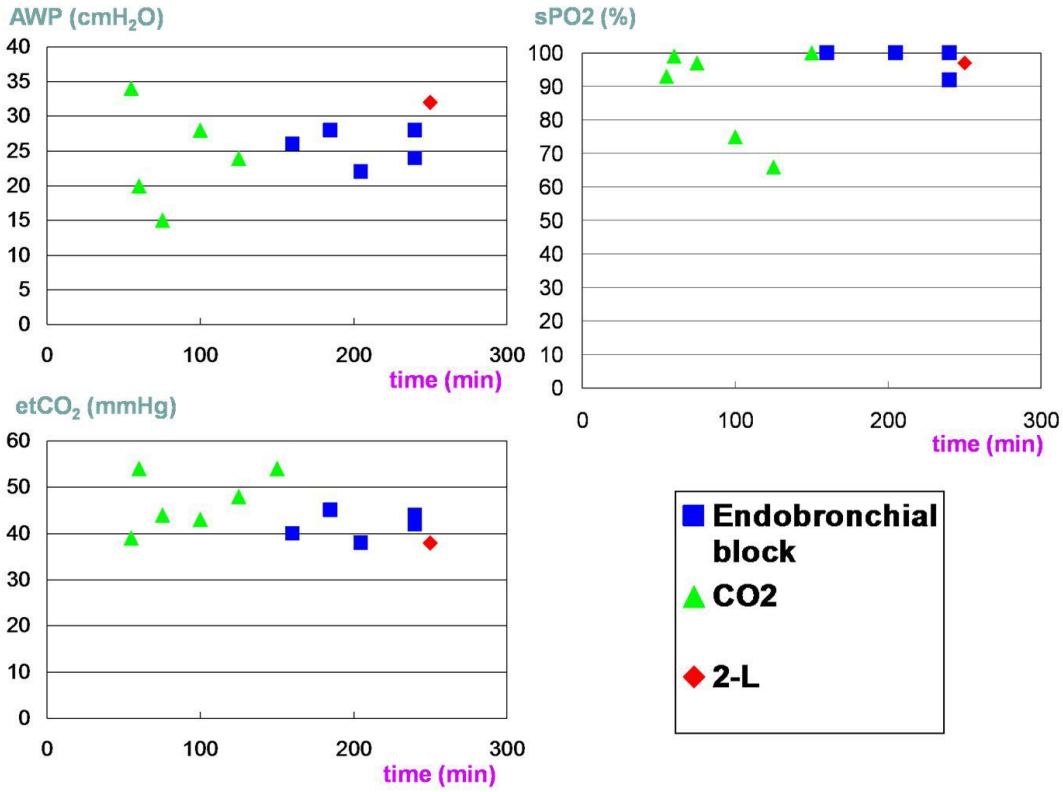

Figure 3: One-lung ventilation methods and time (in minutes) versus important physiological parameters are plotted. AWP: airway pressure

Shows the physiological changes assorted by different one-lung methods and one-lung ventilation time. Endobronchial blocker revealed stable levels of sPO_2, $etCO_2$ and intra-operative airway pressure with less fluctuation.

Discussion

With increasing popularity of prenatal ultrasound, most congenital lung malformations can be detected before birth, and up to 85% of the originally asymptomatic patients became symptomatic after neonatal period [22,23]. The surgical timing for these patients had no formal consensus, and some authors even encourage non-operational watchful waiting for patients without symptoms [24,25]. However, for neonates and small infants, symptoms related to lung lesion may not be limited to breathing conditions or pulmonary infections, but poor feeding tolerance or body weight gain as well. Up to 26% of prenatally diagnosed lesions can have occult infection, cell dysplasia or even tumour
[26]. In addition, few congenital lung anomalies other than CPAM may have similar appearance in prenatal or postnatal images, which will lead to different disease progression [27-29]. No single reliable indicator was found, that is, which asymptomatic patient would remain asymptomatic throughout his/her life span and which patient would develop symptoms in the watchful waiting period.

Most current opinions still propose the performance of surgery in patients' first year [1,2,30]. This study advocates patients to have VATS surgery between 3 and 9 months if they have no breathing conditions. A high rate of pre-operative symptom was observed in the thoracotomy group, which was due to the retrospective nature of our study and selection bias. In this study, VATS surgery, either multiportal or uniportal, cannot be performed smoothly before the age of 3 months or with body weight less than 5 kg due to poor compliance of one-lung ventilation. In the recent two decades, thoracoscopic surgery for paediatric patients has become popular worldwide and even has become a routine procedure for congenital lung malformations [2,6,30]. Meta-analysis reports and retrospective comparative studies indicate that VATS is associated with few complications and short hospital stay [4,6,31,32]. Therefore, the procedure has been proven safe and feasible. Difficulty in such surgical technique is still a problem because low patient numbers in most hospitals inhibit quick accumulation of experiences. Long learning curve must be overcome, and some reports have shown stable results after the first 33 to 50 cases of VATS resection [33,34].

Before 2016, no available English reports were available on uniportal VATS resection for congenital lung malformation in children. Such procedures are mostly performed for spontaneous

pneumothorax, lung wedge resection, intrathoracic biopsy, tumour excision and thoracic empyema [10,12]. The first reported case was an extralobar pulmonary sequestration removed from a 3-cm-long incision of a 14-month-old patient [11]. The largest case series, 19 patients, thus far was reported by Park in 2017 [33]. Uniportal VATS is generally considered more technically challenging than multiportal VATS. Modification has been proposed to prevent instrumental clash by separated ICS incisions of camera and working instruments through a single skin incision [14]. However, the method may increase the risks of over-stretching ribs and intercostal muscles, which may compromise the intention of preventing chest wall deformity by minimal invasive surgery. In this study, initially high conversion rates in multiportal VATS group mainly attributed to the lack of working space. By contrast, uniportal VATS has an advantage by initially creating a relatively large incision for all types of instrument facilitation, especially for tying vessel and endoscopic stapler use. Hence, bleeding control and parenchyma resection can be performed easily without stretching wound or spreading ribs. The complication rates of VATS resection in children differ in the literature, ranging from 4.4% to 30.6% [30,32]. In this study, the VATS complication rate is 38.5% (5/13). Among them, three patients in multiportal VATS group converted to open thoracotomy, and no post-operative or delayed complications developed later. In uniportal VATS group, one patient developed subglottic stenosis, which was related to urgent intubation in ICU rather than surgery. The only true surgical complication was delayed pneumothorax for a CPAM 15-year-old girl and the reported rate of pneumothorax after lung-sparing resection is approximately 10% [35,36]. Blood loss was severe in the multiportal VATS group, and the difficulty of checking and lacking space was the main contributor to the result. In the latest case of VATS lobectomy, the problems seemed to be resolved by endobronchial blocker usage. Long ICU stay, and hospital stay were also found in the multiportal VATS group. Two small patients (4.9 kg and 2.8 kg) had long ICU stay because of prolonged intubation necessity, which may not be directly related to surgical or anaesthesia methods but to lung condition and prematurity. Less analgesic shots were required in the multiportal VATS group; however, the gap was only slightly more than one shot over 7 to 14 days of hospital stay.

For infants and small children, many biological and physiological conditions are different from adults, with small airway and large proportion of dead space of lungs, subsequently decreased gas exchange [37]. Besides, high absorption surface per unit of body weight, less pleural fat and thin vessel walls would increase and accelerate CO_2 absorption and diffusion, which would lead to respiratory acidosis and alternation of cerebral circulation [38]. In haemodynamic and thermal regulations, infants have low tolerance of dramatic changes, which will occur if the early signs of compensation are not identified. Endobronchial blockers for one-lung ventilation showed superior results of stable biological parameters even after a long period of one-lung ventilation during anaesthesia [39,40]. Fibre-optic bronchoscopy-assisted coaxial placement via regular non-cuffed endotracheal tube is proven to be a simple and feasible method even in small infants of 3 months of age or 5 kg of body weight. With such ventilation aid, uniportal VATS can be performed smoothly without the need of conversion. The retrospective nature of this study and patient selection bias may influence the outcome. Apparently, in patients born with severe respiratory symptoms, open thoracotomy is the main choice of surgery. Small patient number is another issue, which may cause under- or overestimation of statistical results. After the initial learning curve, shorter operative time, short hospital stay, and low complication rate will be expected in the future.

Conclusions

Thoracoscopic surgery in paediatrics is feasible, effective and safe under experienced paediatric surgeons with good cooperation from well-equipped anaesthesia team. Uniportal VATS with endobronchial blocker is a preferred surgical approach for small children or infants with congenital lung anomalies. For large patients, multiportal VATS and uniportal VATS can be used smoothly.

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