

Thoracoscopic Lobectomy for Congenital Cystic Lung Disease in Infants

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Abstract

Background: Congenital cystic lung disease is a congenital disease with abnormal development of the trachea, pulmonary parenchyma and pulmonary vessels. Congenital cystic adenomatoid malformation and pulmonary sequestration are the two main types. With the development of endoscopic technology, congenital pulmonary dysplasia has been alleviated in an increasing number of infants through thoracoscopic surgery. However, the timing of the operation is controversial.

Methods: Seventy-two infants with congenital cystic lung disease who underwent thoracoscopic lobectomy from March 2017 to April 2020 were selected as subjects. The data were summarized and analysed, including preoperative complications, operative situation, postoperative recovery, postoperative hospital stay time, etc. All children were followed regularly.

Results: Seventy-two patients were aged 3 to 7 months, with an average age of 5.40 ± 2.02 months: 44 were male, and 28 were female, with an average weight of 6.95 ± 1.96 kg. Preoperative condition: 48 patients were observed and operated on when they were between 5 and 7 months old. Twenty-four patients were diagnosed with pneumonia, and thoracoscopic surgery was performed after anti-infective therapy. All children were diagnosed with congenital cystic pulmonary disease with a CT scan before the operation. Postoperative pathological diagnosis showed 50 cases of congenital cystic adenomatoid malformation, 16 cases of intralobar pulmonary sequestration, and 6 cases of both congenital cystic adenomatoid malformation and intralobar pulmonary sequestration. Anaesthesia was performed by artificial pneumothorax or bronchial occlusion. The average operative time was 92.17 ± 19.65 min, and the average length of postoperative ventilator assistance was 28.18 ± 11.01 hours. Complications: 3 cases of pneumothorax after the operation and 5 cases of atelectasis, and the incidence of pneumonia was 33%. No bleeding, bronchial pleural fistula or other serious complications were observed. All children were followed for 3 months to 2 years. The growth and development of all the children were not different from those of normal children.

Conclusions: In this study, preoperative pneumonia accounted for 23.6% of children. Thoracoscopic lobectomy was effective and feasible for children with congenital cystic lung disease from 3 to 7 months of age.

Background

Congenital cystic lung disease (CCLD) is a kind of congenital disease with abnormal development of the trachea, lung parenchyma and pulmonary vasculature; congenital cystic adenomatoid malformation (CCAM) and bronchopulmonary sequestration (BPS) are the two main types of lesions [1, 2]. CCAM and BPS have similar clinical manifestations, imaging features and histopathological changes[3]. Most cases are diagnosed by ultrasound during pregnancy[4]. After birth, some children are diagnosed with recurrent pneumonia or even respiratory failure. At present, for CCLD, several articles have been reported on thoracoscopic lobectomy in children, but there are few articles on thoracoscopic surgery in children under

one year of age, and the timing of the operation is controversial. This paper summarizes the clinical data of 72 infants with CCLD who received thoracoscopic lobectomy in our hospital and analyses their preoperative condition, intraoperative situation and postoperative recovery.

Methods

1. Clinical data: Retrospective analysis of the clinical data of 72 infants in our hospital from March 2017 to April 2020 (Table 1).

Table 1
Baseline data of 72 infants.

Males	44 (61.1%)
Females	28 (38.9%)
Age (months)	5.40 ± 2.02
Weight (kg)	6.95 ± 1.96
Preoperative pneumonia	24 (33.3%)
CCAM	50
BPS	16
CCAM and BPS	6
Lesion	8
Left upper lobe	
Left lower lobe	20
Right upper lobe	11
Right middle lobe	5
Right lower lobe	25
Both right upper lobe and right middle lobe	3

2. Surgical Methods

Anaesthesia was achieved with endotracheal intubation, and bronchial embolization or piezotreatment was performed. Children were placed lying on the healthy side. One 5-mm trocar was positioned in the 8th intercostal space of the midaxillary line as the observation trocar, another 5-mm trocar was positioned in the 7th intercostal region of the posterior axillary line, and a 3-mm trocar was positioned in the 5th intercostal region of the anterior axillary line. The pneumothorax pressure was maintained at 4–8 mmHg with a flow of 1 L/min and was adjusted according to the degree of collapse of the lung. Pulmonary arteries, veins and bronchi were dissected one by one, the blood vessels were fully exposed, and the

incomplete fissure was cut off with an ultrasonic scalpel. For BPS, lobectomy was performed after disconnection of the isolated pulmonary artery. After dissecting the pulmonary artery and pulmonary vein, the 8th intercostal incision was enlarged to 12 mm, the trachea of the dysplastic lobe was cut off with an endovascular gastrointestinal anastomosis stapler (ENDO-GIA), and the specimen was removed from the 8th intercostal cavity after being placed into a specimen bag. After lung recruitment and if no air leakage was detected, a closed thoracic drainage tube was indwelled, and the thoracic cavity was closed.

Statistical analysis

Data were analysed with the SPSS 18.0 software program (Statistical Package for Social Sciences, SPSS Inc., Chicago, IL, USA). Continuous variables are presented as median values with ranges.

Results

Intraoperative conditions: artificial pneumothorax was established in 51 patients, there were 21 cases of single-lung ventilation, and all lobectomies were successfully completed. Seventy-one cases were completed through three-hole thoracoscopic surgery, and 1 case was assisted by another small incision because of extensive pleural adhesion. Postoperative pathology confirmed 50 cases of CCAM, 16 cases of BPS, and 6 cases of CCAM combined with BPS. The length of the incision was 1.65 ± 0.30 cm, the average operative time was 92.17 ± 19.65 min, and the intraoperative blood loss was 17.06 ± 12.70 ml.

Postoperative conditions: The drainage tube removal time was 6.40 ± 2.76 d, the average length of postoperative ventilator assistance was 28.18 ± 11.01 h, and the postoperative hospital stay time was 8.53 ± 2.11 days.

Complications: Postoperative pneumothorax occurred in 3 cases and atelectasis in 5 cases, and the incidence of pulmonary infection was 33%. No haemorrhage, tracheal pleural fistula or other serious complications were observed, and all the children were discharged successfully. All the children were followed for 3 months to 2 years. During the follow-up, all the children recovered well. The re-examination of lung CT indicated a good recovery, and the growth, development and activity levels were no different from those of the normal children.

Discussion

Are CCAM and BPS treated surgically? At present, most people believe that CCLD in infants may lead to repeated pulmonary infection and respiratory distress and should be removed by surgery as soon as possible [5]. Clinically, it is generally believed that early or emergency surgery should be performed for those with early symptoms, and selective surgery should be performed for those with no complications. Tsai et al. [6] performed surgical treatment on 105 infants, and the incidence of postoperative pneumothorax was 2.9%. In our case, 24 patients had recurrent pneumonia, and intraoperative pleural adhesion and hilar lymph node enlargement were found in those 24 patients, which caused certain

difficulties in dissecting the hilar blood vessels, and old purulent secretions could be seen in the resected lobes.

The timing of surgery for asymptomatic children is controversial. Aziz et al. [7] believe that CCAM may be self-regressive and that infant surgery is risky, so conservative observation should be performed. Eber[8] recommends that in CCLD, pneumonia should be treated early and that surgery should be performed before pneumonia develops. Calvert et al. [9] chose to operate at the age of 3–6 months, which could not only avoid risks such as those associated with surgical anaesthesia in the neonatal period but also avoid long-term complications. We believe that for CCAM and BPS diagnosed by foetal ultrasound, the size and changing nature of lesions monitored by prenatal ultrasound should be regularly reviewed, and postpartum diagnosis should be confirmed by X-ray and CT [10]. After birth, emergency surgery or selective surgery should be decided according to whether the baby has complications and the severity of symptoms. For asymptomatic infants, outpatient follow-up visits should be conducted for regular X-ray and CT re-examination until the age of 3–6 months.

Thoracoscopic lobectomy in children was first reported by Rothenberg[11] in 2000 and has become extensive worldwide in recent years; recent reports have been in older children, and there are few reports of thoracoscopic lobectomy within the first year of life. Due to an infant's narrow chest space, intercostal stenosis, difficulty with one-lung ventilation and other factors, the operation difficulty of infant thoracoscopic lobectomy is increased, which requires the performer to have better operational ability. In our study, 72 infants, with an average operating age of 5.04 months, were all operated on successfully by thoracoscopy.

Conclusions

1. Thoracoscopic lobectomy is effective and feasible for children 3 to 7 months of age with congenital cystic lung disease.
2. According to our data, preoperative pneumonia accounted for 23.6% of children.
3. For extensive pleural adhesion, another assisted incision is more effective and safer for infants.

Abbreviations

CCLD

congenital cystic lung disease; CCAM:congenital cystic adenomatoid malformation;
BPS:bronchopulmonary sequestration; CT:computerized tomography; ENDO-GIA:endovascular
gastrointestinal anastomosis stapler.

Declarations

- Ethics approval and consent to participate:The hospital ethics committee approval was granted of this article.

- Consent for publication was obtained from the child's parents.
- Availability of data and materials: Written informed consent was obtained from the patient for publication of this article.
- Competing interests: The authors declare that they have no competing interests.
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- Authors' contributions: Jinxi Huang participated in clinical practice, contributed to collection and analysis of data, drafting the manuscript, and revising it. Junjie Hong carried out data collection. Qiang Chen participated in clinical practice. Dianming Wu and Songming Hong helped in design of study and drafting the manuscript. Chaoming Zhou carried out patient recruitment and clinical practice, contributed to conception, design, drafting the manuscript, and revising it. All authors read and approved the final manuscript.
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