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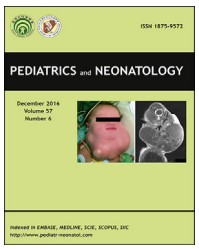

ORIGINAL ARTICLE

Congenital Lung Malformations: Shifting from Open to Thoracoscopic Surgery

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Background: Over the years the need for surgical treatment, timing of intervention, and the type of surgical approach have been discussed, but the treatment of congenital lung malformations remains controversial. The aim of this study was to compare the thoracotomy approach with the thoracoscopic technique by evaluating different surgical outcomes (duration of surgery, postoperative hospital stay, and complications).
Methods: All patients operated from January 2011 to March 2015 for suspected congenital cystic lung were included in the study. Patients treated for congenital lobar emphysema and tracheobronchial neoplasms were excluded from the study.
Results: In the analyzed period, 31 asymptomatic patients were treated: 18 lung resections were performed with thoracotomy (Group A) and 13 with the thoracoscopic approach (Group B). No significant differences were observed between the age and weight at surgery, length of the procedures, complications, and the need for postoperative intensive care between the two groups. The postoperative hospital stay in Group A was twice that for Group 2 ($p = 0.0009$).
Conclusion: Comparing thoracoscopic surgery with the traditional open approach, we confirmed the superiority of minimally invasive treatment in terms of postoperative hospital stay. Common technical recommendations can help pediatric centers to develop the thoracoscopic approach for the treatment of congenital pulmonary malformations.

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1. Introduction

Congenital pulmonary adenomatoid malformations (CPAM) and pulmonary sequestration are the most common congenital pulmonary malformations, with an incidence of 1:10,000 to 1:35,000 newborns, without predilection for side, sex, or race.[1] The treatment of these congenital malformations is one of the most debated topics in pediatric surgery. Surgical resection or active surveillance has been discussed. Three issues have been identified in favor of surgical treatment: (1) risk of malignancy: the differential diagnosis between a lung malformation and pleuropulmonary blastoma (PB) is not always easy on imaging, and lung malformations can transform later into malignant tumors, as reported in literature[2]; (2) prevention of infections, occurring in 10% within the 1st 12 months and in 86% within the 1st 2 years of age according to literature[3,4]; and (3) the lack of validated recommendations on the need and timing of long-term follow-up for those patients who are conservatively treated.[2] The timing of surgery is another matter for debate. Despite the association between lung infections and poor surgical outcome,[5] the timing of surgical treatment has still not been clearly defined.

The thoracoscopic approach was shown to be superior when compared with the open approach in terms of length of postoperative ventilation and hospital stay, as well as the incidence of surgically-related complications.[6] The perioperative anesthetic management is a mandatory part of the overall patient management and it can have a significant impact on the surgical approach. In particular, intraoperative one-lung-ventilation is essential for the adequate visualization and exposure of the other lung, especially during the thoracoscopic approach. This can be challenging, even in tertiary level pediatric institutions, especially for very young patients.[7] Moreover, most devices available on the market for one-lung-ventilation are standardized for adults and no devices are available in infant size.[8] This makes adequate one-lung-ventilation very challenging if surgery is required at an early age.[7,9,10]

The decision to prospectively perform thoracoscopic approach in children with suspected congenital pulmonary malformation (SCPM) was made in 2012 after the adoption of a single-lung ventilation technique with an endobronchial blocker in a case series of 17 patients.[9]

This study reports our initial experience on thoracoscopic resection for pediatric pulmonary malformations, analyzing the differences between an open approach and thoracoscopic resection in terms of surgical outcome.

2. Methods

All children who underwent pulmonary resection for SCPM between January 2011 and March 2015 were retrospectively reviewed. We excluded all the other indications such as primary tracheobronchial tumors or congenital lobar emphysema from the study. Patient data included sex, age and weight at surgery, timing of diagnostic imaging, previous history of pneumonia or respiratory distress, anesthesia management, the American Society of Anesthesiology score (ASA), type of surgical resection (lobectomy or atypical resection) performed, and histology using Stocker classification.[11]

Sparing-lung resection was performed when preoperative computed tomography (CT) scan and intraoperative finding showed only a segmental lesion in order to achieve the parenchymal preservation. This policy is in line with current literature, reporting segmental resection as a safe procedure without risk of residual disease and recurrence.[12,13]

Surgical outcomes included operative time, need of postoperative intensive care unit (ICU), complications, and length of hospital stay. Surgical complications were recorded using Clavien—Dindo classification.[14]

Two groups were identified: Group A (thoracotomy approach) and Group B (thoracoscopy approach). Statistical analysis was performed using Mann—Whitney U test for continuous variables (age and weight at intervention, operative time, length of hospital stay, and ASA score) and Fisher exact test for categorical variables (complications, postoperative ICU), and a p value <0.05 was considered as statistically significant. A 30-day postoperative clinical and radiological (thoracic x-ray) follow-up was performed by the institutional pulmonologist.

3. Results

Between January 2011 and March 2015, a total of 31 pulmonary resections were performed for SCPM with a total of 18 thoracotomies (Group A), and 13 thoracoscopic procedures (Group B). CT was performed at median age of 6 months in both groups (Group A range: 4 days to 5 years; Group B: 2 months to 5 years). All procedures were performed by the same surgeon. All cases of pulmonary sequestration detected with a CT scan were treated with endovascular embolization of the aberrant artery before surgical procedure in order to reduce surgical risk of bleeding and abolish existing shunt.

Of the 18 thoracotomies (Group A), nine were lobectomy and nine atypical resection. A total of eight lobectomies and five atypical resections were performed by the thoracoscopic approach (Group B). Thoracotomies were performed with a muscle-sparing antero-lateral access and anesthesia was conducted with double lung ventilation. Single lung ventilation was obtained with an Arndt 5 French pediatric bronchial blocker (Cook Critical Care Bloomington, IN, USA). Parenchimal resection was performed using Ligasure (ValleyLab Inc., Boulder, Colorado). Vascular and bronchus closure was sutured using ligature or stapler. The surgical specimen was extracted enlarging one

thoracoscopic access. Operative time and demographics are reported in Table 1.

Twenty four-hours postoperative recovery in ICU was required in 12 of 18 patients of Group A and in four of 13 patients of Group B. No statistically significant differences were observed between Group A and Group B in terms of age and weight at intervention, duration of procedure, need for ICU, or ASA score (Table 1). The length of hospital stay was shorter in Group B, with a median stay of 9.5 days for Group A (range: 7–11 days) and 4 days for Group B (range: 4–7 days; $p = 0.0009$).

Twelve CPAM (one Type 1 and 11 Type 2), one PB (local complete resection), and five cases of pulmonary sequestration (three intralobar and two extralobar) were resected in Group A; 11 CPAM (one Type 1 and 10 Type 2) and two cases of pulmonary extralobar sequestration were resected in Group B. All diagnoses were histologically confirmed and all pulmonary lesions were analyzed by the pathologist with evidence of safe margins with healthy lung tissue. After atypical resection, the lung was reventilated by the anesthesiologist under thoracoscopic view to avoid macroscopical residual tissue.

Two patients (Group A) had a Grade 3 Clavien–Dindo complication (one postoperative hemorrhage and one pneumopericardium with surgical reinterventions), but there was no statistical difference between the two groups ($p = 1$, Fisher's Exact test). Neither anesthetic complication nor blocker displacement occurred in any cases.

4. Discussion

Surgical management of congenital pulmonary malformation is a highly contested issue in pediatric patients. Despite several studies, there is lack of a clear indication as to the choice of surgical approach in literature. Our Institution adopted a surgical approach in all patients with SCPM, supporting the current evidence in favor of surgery, based on the controversial diagnosis or development of malignancies.[2] In our patients with PB, the differential diagnosis between PB and benign lung malformation was not possible at preoperative CT (Figure 1). In fact, pathological examination of congenital pulmonary malformation is required to differentiate CPAM from a PB,[15] and Bisogno et al[16] reported the role of a complete surgical resection of PB as a significant favorable prognostic factor.

Surgical management of pulmonary sequestration remains controversial in literature. Although some authors have suggested endovascular treatment as a conservative approach for pulmonary sequestration, reserving surgery only in case of infection, we agree with Cho and colleagues[17] on performing surgery in order to obtain a pathological diagnosis and avoid risk of malignancies.[17–19]

The thoracoscopic approach for congenital lung malformations seems to be a technique gaining acceptance worldwide, although the authors note that Italy is an exception with few case reports.[13,20] The current absence of evidence-based randomized studies does not help pediatric surgeons and pulmonologists that want to start with the thoracoscopic approach of lung malformations.

In literature there is only one prospective study about the application of the thoracoscopic approach presenting a complication rate of 12.5% and conversion of 12.5%.[21] The most relevant series reported an incidence of conversion to open approach ranging from 15% to 33% and identified previous history of pneumonia as the main risk factor to conversion.[5,22,23]

The decision to shift our surgical approach to thoracoscopy was taken on the belief that this technique represented a less invasive procedure in terms of quality of life for pediatric patients with the same results in term of complications. In our two groups, without differences in terms of age, weight, and ASA score at surgery, the most relevant difference concerned the length of hospital stay, which was twice as long in the thoracotomy.

Comparing our results with the only prospective study reported in literature,[21] we did not experience complications and conversion in the thoracoscopic group, but our series differs from Kaneko et al's[21] in two important aspects: adoption of single lung ventilation and different age at intervention.

Table 1 Descriptive statistics of the two groups of patients.

	Thoracotomy ($N = 18$)	Thoracoscopy ($N = 13$)	p^*
	Median (first to third quartile)	Median (first to third quartile)	
Duration (min)	75 (70–95)	105 (75–110)	0.32
Age (y)	1.5 (0.6–4)	1 (0.6–4.1)	0.84
Inpatient stay (d)	9.5 (7–11)	4 (4–5)	0.0009
Weight (kg)	10.5 (8–15)	9.8 (9–18)	1.00
ASA score	2 (2–2)	2 (2–2)	0.80

ASA = American Society of Anesthesiologists.
* p values refer to the Mann–Whitney U test.

Figure 1 Pleuropulmonary blastoma features on computed tomography scan (axial section). Pulmonary tumor presented with macroscopic cyst of right anterior basal lobe and left mediastinal shift.

In the multivariate analysis reported by Seong et al,[22] the age at presentation was not considered a risk factor for conversion or complication. This consideration suggests that the single lung ventilation may represent a key to a prospective adoption of thoracoscopic approach for congenital lung malformations without conversion and complications in early application. In our opinion, the role of anesthesia management is the key to the thoracoscopic approach for congenital lung malformations. Anesthetic time was equally important to the surgical procedure, as an adequate ventilation supports children through major demolitive thoracic surgery and provides the surgeon optimal tissue exposure. In our initial experience of thoracoscopic lung resection for congenital disease, single lung ventilation with endobronchial blocker is the key factor in preventing conversion or complications.

This study has several limitations. Firstly, there was a bias in terms of surgical experience: thoracoscopic approach was started after only a preliminary experience of thoracotomy surgery. Secondly, this prospective study was not randomized, since after the introduction of single lung ventilation, all patients with SCPM were treated with the thoracoscopic approach.

5. Conclusion

In the case of asymptomatic patients, careful monitoring with the first CT after first 6 months provides better surgical results. Surgery provides a definitive diagnosis of SCPM to exclude primary lung tumors.

Comparing minimally invasive surgery with the open approach, we confirmed the superiority of thoracoscopic surgery in terms of postoperative length of hospitalization. Common technical recommendations may help pediatric centers to develop the thoracoscopic approach for congenital lung malformations.

Conflicts of interest

The authors declare no conflicts of interest.

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