Journal of Pediatric Surgery (2013) **48**, 1476–1480

ELSEVIER

Journal of Pediatric Surgery

www.elsevier.com/locate/jpedsurg

Sparing-lung surgery for the treatment of congenital lung malformations ☆

Francesco Fascetti-Leon*, Dalia Gobbi, Stefania Valentina Pavia, Antonio Aquino, Giovanni Ruggeri, Giulio Gregori, Mario Lima

Department of Paediatric Surgery, Paediatric Surgery Unit, University of Bologna, Via Massarenti, 11, 40138, Bologna, Italy

Received 4 July 2012; revised 16 January 2013; accepted 20 February 2013

Key words:
CCAM;
Congenital cystic lung;
Thoracoscopy;
Segmental resection;
Sparing-lung, wedge resection

Abstract

Purpose: Congenital lung malformations (CLM) are traditionally treated by pulmonary lobectomy. The foremost indication for surgery is that these lesions predispose to respiratory tract infections and to malignancy. Owing to the improvement of prenatal diagnosis, most patients are operated in a timely manner and prophylactically. In this context, lung-sparing surgery (LS) has gained interest as a lung preservation strategy, especially for asymptomatic patients. In the present study, we evaluated both thoracoscopic and open lung-preserving resections as an alternative to lobectomy for CLM resection.

Methods: We retrospectively reviewed all patients who underwent lung-sparing resection (segmentectomy and atypical resection) for CLM from 2001 to 2010. Data were collected regarding preoperative diagnostic workup, type of intervention, and follow up.

Results: Fifty-four patients received LS for CLM during the study period. Twenty-six were approached thoracoscopically, with 18 cases requiring open conversion for a complete resection. There were six postoperative complications: three asymptomatic pneumothoraces that resolved without intervention, one tension pneumothorax that required replacement of a drainage catheter, and three instances of intraoperative bleeding requiring blood transfusion. Mean duration of follow-up was 65.2 months. Two patients experienced pneumonia during the follow-up period. A third patient had a cystic lung lesion on postoperative computed tomography (CT) which required a second-look surgery.

Conclusions: LS for CLM is a safe and effective means of lung parenchymal preservation in pediatric patients. Complication rates are comparable to that of traditional lobectomy. In our experience, this type of lung surgery does not carry a higher risk of residual disease and recurrence if accurately planned in selected patients, i.e., those with small asymptomatic lesions. The complication rate is acceptable and apparently not affected by preoperative symptoms. The thoracoscopic approach is recommended, although open conversion should be advocated to avoid too long operative times.

☆ The authors declare that no conflict of interest exists regarding the paper and give the final approval of this version to be published.

* Corresponding author. Pediatric Surgery Unit, University of Bologna, Via Massarenti 11, 40138 Bologna, Italy. Tel.: +39 3283655620; fax: +39 051 6364657.

E-mail addresses: f.fascettileon@gmail.com (F. Fascetti-Leon), daliahg@yahoo.com (D. Gobbi), stefaniapavia@hotmail.com (S.V. Pavia), aquinoantonia@virgilio.it (A. Aquino), giovanni_ruggeri@aosp.bo.it (G. Ruggeri), giulio.gregori@hotmail.com (G. Gregori), mario.lima@unibo.it (M. Lima).

0022-3468/$ – see front matter © 2013 Published by Elsevier Inc.
http://dx.doi.org/10.1016/j.jpedsurg.2013.02.098

The spectrum of congenital lung malformations (CLMs), elsewhere named congenital thoracic malformations, includes congenital adenomatoid cystic malformation (CCAM), bronchopulmonary sequestration, bronchogenic and foregut cysts, bronchial atresia with distal cystic degeneration, and congenital lobar emphysema. All these lesions can be seen during prenatal assessment but definitively distinguished from one another after histological examination [1,2].

There is a wide acceptance that postnatal symptomatic lesions (i.e. infection, hyperinflation, pneumothorax, left to right shunting, and pulmonary hypoplasia due to large mass) require surgical resection [3]. On the other hand, the management of asymptomatic lung lesions is controversial because of concerns related to operative morbidity and mortality, and to the uncertain outcome because of lack of long term follow-up [4–7]. Improvements in prenatal imaging have enhanced the therapeutic dilemma of these infants. The risk of respiratory failure, recurrent respiratory tract infections, and airway obstruction, and reports of malignant degeneration in CCAM (bronchioloalveolar carcinoma, rhabdomyosarcoma, pleuropulmonary blastoma) [6–8], have further complicated the decision-making process, and today represent the foremost indications for surgical resection. Herein we present a series of non-anatomical lung resections for CLMs performed in a single center in a ten-year-period. The goal of the study is to examine the efficacy and the morbidity associated with lung-sparing (LS) surgery as a possible, definitive treatment of CLMs. The second aim was to assess the role of mini-invasive surgery for this disease in our institution.

1. Methods

In the present study we retrospectively reviewed the records of the patients, referred to the Pediatric Surgery Department of Bologna University, who underwent atypical pulmonary resection for CLM between January 2001 and December 2010. The study complies with the Declaration of Helsinki and with the Italian Law Decree n.196/2003 to protect personal data. Data collected from charts included age at operation, sex, prenatal diagnosis, preoperative symptoms, pathologic diagnosis, type of procedure performed, length of hospital stay, time of chest tube in place, postoperative complications, postoperative clinical and radiological results at follow up. All these patients underwent clinical follow up, a radiological evaluation with a chest radiograph 1 year after surgery, and a chest computed tomographic (CT) scan when the chest radiograph or symptoms raised the suspect of residual disease. Data were analysed using the chi-square test Yates-corrected and Student's t-test. Values are expressed as a mean ± standard deviation (SD).

2. Results

A total of 81 patients underwent intra-thoracic resection for CLM during the study period. Fifty-four received LS surgery. Table 1 reports the patients' characteristics. Age at the time of surgery ranged from 16 days to 14 years (mean 8.6 months). A preoperative CT scan was performed in all patients at the average age of 3.2 months; multiplanar reconstruction has been used constantly but became essential in those CT cases with the deeper lesions (Figs. 1, 2). Forty-seven patients (92%) had prenatal diagnosis between 20 and 32 weeks' gestation. Nine were symptomatic before surgery: 6 presented with neonatal respiratory distress, 3 with bacterial pneumonia (at 15, 48 and 60 months respectively). 46 patients (85%) received a formal muscle sparing thoracotomy (28 primary, 18 conversion after thoracoscopy). Twenty-six of 54 had an initial thoracoscopic approach (48%) that was successfully completed in 8 cases (31%). Lobar involvement, type of lung resection and histological diagnoses were specified in Tables 1 and 2. In all cases histopathological studies demonstrated safe margins. Mean operative time was 122.2 min (ranged from 65 to 255 min). In all cases lung tissue was transected along non-anatomical segmental lines following the lesion edges (visible, palpable or radiographically detected). Four patients underwent anatomical segmentectomies because these lesions were completely contained in a segment with safe margins. Tissue division was adequately carried out combining the use of LigaSure device, bipolar cautery, or endoscopic stapler. Absorbable running suture of the edges was applied in case of wide parenchymal resection to seal the section plane. A postoperative chest tube was positioned in all patients for a mean time of 5.5 days (1–17 days). The interventions were accomplished by two surgeons. Three patients experienced intra-operative bleeding requiring transfusion. Three patients had persistent but asymptomatic pneumothorax that hampered chest tube removal; one had a tension pneumothorax at tube removal that required a second emergent chest tube placement for a total of 27 days. The duration of hospital stay ranged from 7 to 45 days, with a mean of 9 days. Mean duration of follow-up was 65.2 months (range 12–115 months). All patients were followed up with a regular clinical monitoring (outpatient clinic visit at the 1st, 6th and every 12th month) and with radiological evaluation (chest radiograph 3 and 12 months after the operation). Postoperative CT scan was performed only in case of symptoms or persistence of abnormalities on the plain x-ray, and at least 12 weeks after surgery.

After discharge all but 4 patients remained asymptomatic: two had readmission for pneumonia 6 and 36 months after surgery respectively, and the other 2 had recurrent upper airways infections. Other 6 patients had a suspicious image at chest radiograph. Thus, a total of 10 CT scans were done (18.5%) in the follow-up. In the 4 symptomatic patients CT was performed after proper antibiotic therapy. Scarring around resection lines was a common finding, and no lesions