Thoracoscopic surgery for congenital lung lesions—is this the future?

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Congenital pulmonary airway malformations (CPAM) represent the majority of congenital lung lesions (CLL), constituting 30–40% of lung bud developmental anomalies (1). The lesions are benign, and are composed of non-functioning pulmonary tissue; they are usually limited to one lobe. They can be detected in prenatal ultrasound (US) as cystic lesions of variable sizes (2). In 10–20% of cases, additional anomalies can be found (3). The Stocker classification (types 0-IV) is used for CPAMs, and refers to histologic findings and US appearance (4). In up to 65% of cases, there have been reports on spontaneous intrauterine regression (3); however, these lesions are usually visible in fetal magnetic resonance imaging (MRI) or computed tomography (CT) performed after birth (1).

There is a wide variety in the clinical presentation and severity of CPAMs, mostly determined by the degree of lung involvement and the location in the thoracic cavity (5). While the most severe lesions may result in nonimmune fetal hydrops and fetal death, less prominent lesions may present with postnatal complications or even diagnosed incidentally in adult age. The possible postnatal complications are respiratory symptoms, infections and a risk for malignant transformation of the lesion. Many specialized centers are concerned by the malignant potential, and recommend elective surgical resection, even of asymptomatic lesions (6).

Some authors advocate sparing-lung resection when pre-operative CT scan and finding during operation show only a segmental lesion, with the advantage of possible preservation of lung parenchyma (7). However, case reports and small series found that precursors of mucinous bronchioloalveolar carcinoma may be harbored in CPAM type 1; moreover, pleuropulmonary blastoma can be indistinguishable from CPAM type 4. As segmental resection may result in inadequate surgical margins, and carries risk of recurrence, most authors recommend formal lobectomy over segmental resection for CLL (8).

While there is a wide agreement on the need for surgical resection of CLL even in asymptomatic neonates, the timing of operation is still under controversy. It is estimated that 10–30% of asymptomatic neonates will develop an infectious complication before age 1 year; the presence of infection was found to correlate with higher rates of intra- and post-operative complications and longer hospital length of stay (LOS). Additionally, resection at early age offers the theoretical advantage of compensatory lung growth (8). However, in a prospective study with long-term follow-up, age at operation did not correlate with eventual pulmonary function or exercise capacity following lobectomy (9).

The traditional surgical approach has been open thoracotomy through posterolateral thoracotomy. Open thoracic surgery at early age was found to be linked with later chest wall asymmetry, poor cosmetic results and scoliosis (10). In 2003, Rothenberg was the first to report thoracoscopic lung resection in children. His group included 45 patients aged 2 days to 18 years, weighing

2.8 to 78 kg (11). Since then, the thoracoscopic approach has been described as technically challenging, but with reduced short- and long-term morbidity compared to open surgery (10). During the thoracoscopic approach, the visualization and exposure of the operated lung requires one-lung-ventilation. This may pose a technical challenge, especially for very young infants, even in tertiary pediatric centers (7). Over the following years, more experience was achieved in other thoracoscopic procedures, and thoracoscopic resection for CPAM has gained increasing international popularity. In several studies, thoracoscopic lobectomy was found to have advantages over thoracotomy in terms of shorter hospital LOS, less post-operative pain and less long-term musculoskeletal complication (12).

In this regard, we read the interesting paper by Ito *et al.* (13). This is a retrospective study evaluating 13 patients that underwent operation at early age for CPAM. The aim was to evaluate the ideal timing for surgery, and to examine the feasibility of complete video-assisted thoracoscopic surgery (C-VATS). Four infants were operated in the neonatal period (mean age 5 days; range, 1–8 days), due to respiratory failure; all were diagnosed prenatally and underwent open thoracotomy. Nine infants were operated at a mean age of 2 years 3 months (range, 12–68 months); five were diagnosed prenatally and four were operated due to symptoms associated with infection. Four infants underwent C-VATS surgery. Overall, lobectomy was performed in eight cases, lobectomy plus segmentectomy in one case, and partial resections in the other cases. Patient's age and height at operation, operation time, amount of blood loss, time to removal of drain and LOS after surgery were similar between thoracotomy and C-VATS groups, while the body weight was higher in C-VATS group (P=0.03). Thus, for asymptomatic patients, the authors recommend performing surgery after the age of one year. Moreover, they suggest that C-VATS lobectomy may be feasible after the age of 18 months or weight more than 10 kg.

Several other retrospective studies compared the results of open and thoracoscopic surgeries for CLL, with varying results. In a meta-analysis of six retrospective studies, length of hospitalization and time until chest tube removal were longer with the open approach, while the duration of surgery and overall complication rates were similar. Postoperative pain management could not be compared due to heterogeneity between studies (14). Later on, Mattioli *et al.* examined 31 lung resections (13 thoracoscopic). LOS was twice in the thoracotomy group (median 9.5 *vs.* 4 days, P=0.0009) (7). In a large retrospective study of

1,120 children with CLL, 39.4% underwent thoracoscopic surgeries. In 2008, 32.2% procedures were performed thoracoscopically, compared to 48.2% in 2012. The prevalence of open surgeries was higher in newborns, infants with congenital anomalies or comorbidities and those with pulmonary infections. After adjusting for patient complexity and stratifying by the extent of resection, the rate of complications and postoperative LOS were similar between open and thoracoscopic approaches (P=0.061) (10).

The failure of thoracoscopic surgery may necessitate conversion to open procedure. Vu *et al.* found a conversion in 6 out of 18 patients (33%). Patients with respiratory symptoms before operation had a higher incidence of conversion compared to asymptomatic patients (15). Procedures done early in the surgeon learning curve, as well as patient age less than 5 months, were also found to increase the risk for conversion (8). In a prospective study of 45 infants (22 CPAMs), two patients needed conversion to open surgery; none of them had CPAM (16).

There is limited data about the long-term results of open *vs.* thoracoscopic surgery. Recently, Lau *et al.* examined 24 patients, at least 7 years after surgery for CPAM (12 thoracoscopic). Similar to previous findings, children with respiratory infections, co-morbidities and pre-operative respiratory failure were more likely to undergo an open procedure. The thoracoscopic group showed better FVC, FEV1 and diffusion capacity of carbon monoxide, with no statistical difference in lung volumes (12).

Our current practice includes close follow-up of antenatally diagnosed CLL. In some cases, a multidisciplinary meeting is held, involving the treating gynecologist, a pediatric pulmonologist and a pediatric surgeon. The woman is advised to give birth in a tertiary medical center. Soon after birth, the newborn is examined and usually a chest X-ray is performed. If asymptomatic, chest CT is performed around age 9 months, with following surgery. We advise performing lobectomies for CLL. In our medical center, 29 surgeries for CPAM were performed since 2000, with a median LOS of 3 days. In adults, one-lung ventilation with a double lumen tube is less challenging. Additionally, post-thoracotomy pain is more evident in adults compared to the flexible pediatric chest wall. Therefore, most lobectomies in adults are performed thoracoscopically.

In conclusion, thoracoscopic surgery is an emerging technique for asymptomatic CPAMs. The sample size in the study by Ito *et al.* is too small to draw further conclusions, but there is growing evidence that the technique may is feasible and safe, in tertiary centers and dedicated

anesthesiologists. Larger prospective studies are warranted, to define the best patient selection, timing and long-term results of thoracoscopic surgery.

Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at http://dx.doi.org/10.21037/jtd.2020.03.36). The authors have no conflicts of interest to declare.

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