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Optimal timing for elective resection of asymptomatic congenital pulmonary airway malformations

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ABSTRACT

Purpose: We sought to determine optimal timing for CPAM resection within the first year of life.
Methods: We queried the National Surgical Quality Improvement Program pediatric database from 2012 to 2015 for elective CPAM resections on patients less than 1 year of age. Patients were divided by age in months: 1–3 (n = 57), 4–6 (n = 135), and 6–12 (n = 214). Patient operative variables and 30-day postoperative outcomes were compared.
Results: A total of 406 patients were included with no differences in demographics or comorbidities. Median operative time increased with each older age category (115 min, 152 min, 163 min, respectively; $p < 0.01$). Thoracoscopic approach was less utilized in 1–3 months (40.4%) compared to the older two age categories (65.9% and 69.6%, respectively; $p < 0.01$). There were no differences by age in major complications, conversion to open, or readmissions. On multivariate analysis, ASA class≥ 3 ($p < 0.01$) and prolonged operative time ($p < 0.01$) were associated with a major complication. Furthermore, operations on patients aged 6–12 months were associated with increased operative time ($p < 0.01$) regardless of operative approach.
Conclusion: Elective CPAM resections are equally safe in patients 1–12 months of age. Earlier resection including both open and thoracoscopic resection is associated with decreased operative time.
Level of Evidence: IIc, Outcomes Research.

Congenital pulmonary airway malformations (CPAM) are one of the most common congenital lesions in the lower respiratory tract [1–3]. These lesions include congenital cystic adenomatoid malformations (CCAM), extra-pulmonary sequestrations (EPS), intra-pulmonary sequestrations (IPS), and hybrid lesions. They are characterized by abnormal growth of lung tissue leading to the formation of cysts of variable sizes [4,5]. Their incidence is about 1 in 25,000 newborns [5,6]. Detection of lesions usually occurs in utero between 18 and 24 weeks gestational age (GA) [3]. Although there have been many pathological classification systems of CPAM, the most clinically relevant information is the size of the cysts [microcystic (<5 mm) or macrocystic (>5 mm)], the presence of feeding vessels, and association with other lung parenchyma.

Prenatally, large lesions can precipitate isolated polyhydramnios and the development of fetal hydrops. In these settings, fetal therapy is warranted [1,3,7–9]. After birth, the neonate is at risk for respiratory distress or respiratory compromise secondary to mass effect on the functioning lung, necessitating immediate resection. Resection of symptomatic lesions is mandatory [2,3]. If the child remains asymptomatic past the neonatal period, resection is still recommended to avoid emergence of symptoms as has been reaffirmed by a recent meta-analysis [10]. These lesions are prone to infection, may represent an occult malignancy, or may degenerate into malignancy later in life. Pneumonia, pneumothorax, and further complications also threaten the patient if lesions are not resected [2,5,11–13]. Differentiating lesions from normal lung tissue is difficult to do intraoperatively and segmental resections often leave behind parts of lesions, which may require a second resection to remove remaining disease [5]. As a result, lobectomy is preferred over segmental resection in order to resect all CPAMs, excepting EPS. Although thoracotomy was previously necessary for safe resections of a lobe, advances in video-assisted thoracoscopic surgery (VATS) have allowed for successful operations with less morbidity at younger and younger ages [6,14–16].

It is generally held that surgery should be conducted before one year of life to remove the lesion prior to the development of symptoms, restore respiratory function [11,17], and permit compensatory lung growth. Some authors argue for earlier resection within the first few months of life [11,18]. Our study sought to use the National Surgical Quality Improvement Program Pediatric (NSQIP-P) database to determine the postoperative outcomes by age of operation for surgical

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treatment of asymptomatic CPAM. We also looked at operative time as a proxy for technical difficulty with the hypothesis that earlier age would be technically easier, and thus would require shorter operative times, because of smaller vessels, more complete fissures, and less inflammation.

1. Methods

This study was approved by the Johns Hopkins Institutional Review Board. Data from the 2012 to 2015 ACS-NSQIP Pediatric participant user file (PUF) was used. NSQIP-P collects over 90 data points from most pediatric surgical subspecialties with the goal of collecting highly reliable clinical data to compare surgical outcomes across participant hospitals in the program [19]. NSQIP-P samples and collects data in 8 day cycles with 35 procedures per cycle by dedicated clinical abstractors. Demographic information, comorbid diagnoses, laboratory values, operative variables and 30-day outcomes are recorded. Operations are recorded by their primary procedure Current Procedural Terminology (CPT) codes.

Patients were identified using International Classification of Disease, Ninth and Tenth Revision, Clinical modification (ICD-9-CM, ICD-10-CM) diagnostic codes and CPT codes. ICD-9-CM codes used were 748.4 (congenital cystic lung), 748.5 (agenesis, hypoplasia, dysplasia of lung: absence of lung, aplasia, hypoplasia, sequestration), 748.6 (other anomalies of the lung), 748.60 (anomaly of lung, unspecified), 748.69 (other: accessory lung; azygos lobe), 770.2 (interstitial emphysema and related conditions), and 492.8 (Other emphysema NOS). ICD-10-CM codes used were Q33.0 (congenital cystic lobe), Q33.1 (accessory lobe of lung), Q33.2 (sequestration of lung), Q33.8 (other congenital malformations of lung). Patients were selected based on corresponding CPT codes: open wedge or segmentectomy (32,097, 32,484, 32,505), open lobectomy (32,480, 32,486), open resection of two lobes (32482), thoracoscopic wedge resection or segmentectomy (32,608, 32,655, 32,666, 32,669), thoracoscopic lobectomy (32663), thoracoscopic resection of two lobes (32670).

Patients less than 30 days and greater than 1 year were excluded in order to select for asymptomatic elective cases. Children less than 30 days of age undergoing resection were likely symptomatic and possibly emergent. Children older than one year are often incidentally diagnosed after they have become symptomatic at a later age. Patients reported as having an urgent or emergent procedure, or requiring ventilator support within 48 h prior to the operation were also excluded. Age categories were divided into 1 month to 3 months, 4 months to 6 months, and ≥6 months to 1 year. Thoracoscopic procedures were identified if either the primary or associated procedure CPT codes included thoracoscopy. Conversion of thoracoscopic to open procedures were defined as any case with both a thoracoscopic and open procedure CPT code and/or those listed as "Laparoscopic/MIS and Open" category in the NSQIP-P data. Resection status was categorized as wedge, single lobe, or two lobes.

Patient characteristics, comorbidities, operative factors, and 30 day post-operative outcomes were collected and compared across the three age categories. Preoperative comorbidities were categorized by systems: respiratory, congenital cardiac, gastrointestinal, or neurologic. Given the rarity of any single complication, individual complications were grouped into a composite outcome. Major complications included deep incisional or organ space infection, deep wound dehiscence, unplanned reintubation, post-operative pneumonia, pulmonary embolus, renal failure or insufficiency, sepsis, blood transfusion, deep venous thrombosis, post-operative vent requirement over 24 h, reoperation, and new oxygen requirement on discharge. Minor complications include superficial surgical site infection (SSI), superficial wound disruption, and urinary tract infection. Other outcomes included 30-day unplanned readmission, conversion from thoracoscopic to open operation, and postoperative length of stay.

1.1. Statistical analysis

Descriptive findings were presented as median and interquartile ranges for continuous variables and number and percentage for categorical variables. Fischer's exact test or chi-square test was used where appropriate to compare categorical variables across age groups. Kruskal-Wallis test was used to compare continuous variables across age groups. $p < 0.05$ was considered significant. Simple and multivariate logistic regression was used to calculate unadjusted and adjusted odds ratios for binary outcomes. Median simple and multivariate regression was used for continuous outcomes. The factors for the multivariate model were chosen based on significance ($p < 0.05$) of the bivariate analysis for each outcome as well as clinical relevance as determined collectively by the authors. All analysis was performed using STATA version 14.2 (StataCorp 2015, College Station, TX).

2. Results

A total of 406 patients were included in the study, with 57 (14%) in the 1–3 month age group, 135 (33.3%) in the 4–6 month age group, and 214 (51.7%) in the 6–12 month age group. Table 1 describes patient demographic, preoperative, and operative characteristics. No significant difference was found in gender, race, ethnicity, or prematurity. There was a higher percentage of patients requiring oxygen supplementation in the youngest group, but no difference in overall respiratory comorbidity. Cardiac risk factors, gastrointestinal and neurologic comorbidities, as well as preoperative parenteral nutrition, were not significantly different across age groups.

Operative time was found to increase with advancing age with a median of 115 min (IQR 77–160) in the 1–3 month age group, 152 min (IQR 107–215) in the 4–6 month age group, and 163 min (IQR 121–215) in the 6–12 month age group ($p < 0.01$). Anesthesia time, when subtracting the operative time, was not found to be different across age groups. There was no difference in ASA class or surgery type (wedge, single lobe, double lobe). There was a significantly higher rate of thoracoscopic surgery in the older age group (69.6%) compared to the youngest age group (40.4%), $p < 0.01$.

Thirty-day post-operative outcomes as totals and by age category are shown in Table 2. There was no difference overall in minor or major complications across age groups. Within major complications, the most common findings were respiratory, particularly prolonged ventilator requirements >24 h, and blood transfusions. Four patients (7%) in the 1–3 month group, one (0.7%) in the 4–6 month group, and four (1.9%) in the 6–12 month group required ventilator support for more than 24 h.

Table 3 displays unadjusted and adjusted odds ratios for risk factors for a major complication, readmission, and conversion to open, as well as regression coefficients for risk factors for operative time. Age category did not significantly affect the risk of a major complication before or after adjustment. An increase in operative time (OR 1.21; $p = 0.01$) and ASA class ≥3 (OR 2.37; $p = 0.01$) was found to increase the risk of a major complication after adjusting for age, operation type, extent of resection, and respiratory comorbidities. No risk factors were identified for readmission on unadjusted or adjusted analysis. Conversion from thoracoscopic to open was not affected by age category, but increased with operative time (OR 1.21; $p = 0.02$) as would be expected.

Operative time was found to be longer with the 6–12 month age group (median time + 35 min; $p = 0.01$) when compared to the 1–3 month age group after adjustment in the multivariate model ($p = 0.01$). Thoracoscopic operations (+25 min; $p = 0.02$), conversion to open (+37 min; $p = 0.05$), and more extensive resection type (+49 min; $p = 0.00$) were also associated with an increase in operative time.

3. Discussion

Elective resection of asymptomatic CPAMs is advocated because of the risk of infection, pneumothorax, and malignancy [1,2,7]. Furthermore,