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Resection of pediatric lung malformations: National trends in resource utilization & outcomes

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ABSTRACT

Purpose: We sought to determine factors influencing survival and resource utilization in patients undergoing surgical resection of congenital lung malformations (CLM). Additionally, we used propensity score-matched analysis (PSMA) to compare these outcomes for thoracoscopic versus open surgical approaches.

Methods: Kids' Inpatient Database (1997–2009) was used to identify congenital pulmonary airway malformation (CPAM) and pulmonary sequestration (PS) patients undergoing resection. Open and thoracoscopic CPAM resections were compared using PSMA.

Results: 1547 cases comprised the cohort. In-hospital survival was 97%. Mortality was higher in small vs. large hospitals, $p < 0.005$. Survival, pneumothorax (PTX), and thoracoscopic procedure rates were higher, while transfusion rates and length of stay (LOS) were lower, in children ≥3 vs. <3 months ($p < 0.001$). Multivariate analysis demonstrated longer LOS for older patients and Medicaid patients (all $p < 0.005$). Total charges (TC) were higher for Western U.S., older children, and Medicaid patients ($p < 0.02$). PSMA for thoracoscopy vs. thoracotomy in CPAM patients showed no difference in outcomes.

Conclusion: CLM resections have high associated survival. Children <3 months of age had higher rates of thoracotomy, transfusion, and mortality. Socioeconomic status, age, and region were independent indicators for resource utilization. Extent of resection was an independent prognostic indicator for in-hospital survival. On PSMA, thoracoscopic resection does not affect outcomes.

Congenital lung malformations (CLM) are relatively rare with estimates of about 1 in 10,000 to 1 in 35,000 pregnancies [1–5]; however, the incidence has recently risen because of growing recognition of these lesions on the 16–20 week prenatal ultrasound [4–10]. While CLM may regress later in gestation, they may also remain constant, and a small proportion continue to grow resulting in pulmonary hypoplasia of the contralateral lung, mediastinal shift, polyhydramnios, progressive cardiac failure, hydrops fetalis, or intrauterine death [1,9–12].

The most common CLM are congenital pulmonary airway malformations (CPAM), pulmonary sequestration (PS), and mixed/*hybrid* lesions [9]. Although the majority of these lesions will be asymptomatic after birth, infants with CPAM or PS may develop respiratory distress or infections and require surgery [9,13]. Management for the asymptomatic group remains controversial, and if surgery is performed in such patients, there is a lack of consensus regarding the best timing for the procedure as age at resection is thought to affect outcomes [9,16–22].

Further debate has emerged regarding the best surgical procedure and approach for these lesions. While lobectomy is considered the standard procedure for CLM resections, some propose segmentectomy, which can preserve more native lung tissue, as a reasonable alternative in select patients [6,14,19–21,23,24]. Additionally, thoracoscopic resection has been shown to be a safe, feasible operation, which avoids the long-term sequelae such as musculoskeletal complications associated with an open approach [21,25–28]; however, higher risks of perioperative complications in both patients with respiratory distress and in younger, smaller infants may limit use of thoracoscopy in such cases [21,29,30]. Despite the growing body of literature describing the safety and efficacy of the varying surgical techniques, only one study exists, which compares thoracoscopic versus open approaches with propensity score matching to account for variables such as comorbidities and age in the analysis [28].

In addition to debating the risks and approaches of surgical resection, studies have shown that disparate resource utilization and poor

Abbreviations: CLM, congenital lung malformations; PSMA, propensity score-matched analysis; CPAM, congenital pulmonary airway malformation; PS, pulmonary sequestration; PTX, pneumothorax; LOS, length of stay; TC, total charges; KID, Kids' Inpatient Database; HCUP, Healthcare Cost and Utilization Project; AHRQ, Agency for Healthcare Research and Quality; ICD-9-CM, International Classification of Diseases, Ninth Revision, Clinical Modification; USD, United States dollars; MIQ, median household income by zip code quartile; OR, odds ratio; CI, confidence interval; CGH, freestanding children's general hospital; CUGH, children's unit in a general hospital; NCH, non-children's hospital; PFT, pulmonary function tests.

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outcomes in surgical pediatric patients are associated with patient demographics [31–33]. Race, age, and insurance status have been shown to be independent predictors of postoperative mortality, morbidity, length of stay (LOS), and total charges (TC) [32,33]. Furthermore, hospital characteristics have been noted to be associated with these outcome measures in adult and pediatric surgical procedures [34–39]; however, these associations are dependent on procedure type and have not yet been evaluated in CLM patients.

Accordingly, the purpose of our study was two-fold. First, we sought to determine the factors influencing in-hospital survival and resource utilization in patients undergoing surgical resection of CPAM and PS and to evaluate whether patient age at resection was a significant determinant of these outcome measures. In addition, we aimed to determine if a thoracoscopic versus open surgical approach affects these outcomes when accounting for factors such as preoperative comorbidities and age using propensity score matching.

1. Materials and methods

The Kids' Inpatient Database (KID) is a national database available from the Healthcare Cost and Utilization Project (HCUP), sponsored by the Agency for Healthcare Research and Quality (AHRQ). The dataset contains information on approximately 2–3 million pediatric inpatient discharges per triennial release. Clinical diagnoses and procedures are coded using the *International Classification of Diseases*, *Ninth Revision*, *Clinical Modification* (ICD-9-CM). This study included cases from the 1997, 2000, 2003, 2006, and 2009 releases. Data was not collected from intervening years.

Children and adolescents (<20 years old) diagnosed with CLM were identified using codes 748.4 (CPAM) and 748.5 (PS). We limited the cohort to those undergoing surgical intervention for CLM in the form of segmentectomy (32.3, 32.30, 32.39), lobectomy (32.4, 32.41, 32.49), pneumonectomy (32.5, 32.50, 32.59), thoracotomy (34.01, 34.02, 34.99), or other pulmonary resection (32.20, 32.6, 32.9, 33.1), using their respective procedure codes. The procedures were further classified as either thoracoscopic or open surgery. Those with disposition coded as *transfer to short term hospital* and *other transfers, including skilled nursing facility, intermediate care, and other type of facility* were excluded from survival analyses to avoid the potential for duplicate case reporting from the receiving hospital. All analyses were limited to data available for each category and, as such, all analyzed outcomes were limited to the hospital stay associated with the operation.

Categorical measures were compared using chi-square and Fisher's exact tests as appropriate. Continuous measures were compared using Student's *t*- and Mann–Whitney *U* tests as appropriate. Ordinal logistic regression models were constructed to predict demographic, hospital, and clinical characteristics associated with higher resource utilization in the forms of LOS and TC. Binary logistic regression modeling, using a backward-stepwise method, was performed with the same covariates to analyze in-hospital survival characteristics. All analyses were two-sided, and significance was defined at alpha level 0.05. Owing to the nature of the KID, all survival/mortality analyses in our study refer to in-hospital survival/mortality. TC analyses were adjusted to 2009 U.S. dollar (USD) values according to rates determined by the United States (U.S.) Department of Labor [40]. All cases were weighted to project nationally representative estimates.

Propensity score matching to analyze outcomes of thoracoscopic vs. open resection for CPAM was performed using the nearest neighbor 1:1 ratio method. The resulting groups were weighted after matching, thus resulting in unequal numbers in the comparison groups. Briefly, a multivariate logistic regression model was constructed to assign propensity scores for each case included in this analysis. Covariates used for modeling included demographic (age, gender, race), socioeconomic (primary payer, median income quartile), and hospital characteristics (size, location/teaching status, region, type). Risk-adjustment was performed using the comorbidity codes as described in the Elixhauser method, which has been validated in multiple previous studies [32,33,41].

Case matching was performed using *MatchIt* version 2.4–20 (Cambridge, MA), a supplemental module available for *R commander* version 2.14.2 (R foundation for Statistical Computing; Vienna, Austria) [42]. All other statistical analyses were performed using SPSS Statistics version 21 (IBM Corporation, Armonk, NY). The Institutional Review Board at the University of Miami Miller School of Medicine (Miami, FL) deemed this retrospective study to be exempt from full review.

2. Results

A total of 1547 cases of CLM undergoing surgical intervention were identified with an overall in-hospital survival of 97.2%. From 1997 to 2009, the number of surgical CLM cases increased almost three-fold. CPAM accounted for 870 cases (56.2%), and PS accounted for 613 cases (39.6%), while 64 (4.1%) were diagnosed with both. Average LOS was 10.3 ± 20.1 days, while TCs were 72,134.86 ± 121,786.64 USD. Average age at admission was 2.18 years. Additional information regarding cohort characteristics is shown in Table 1.

2.1. Resource utilization

Resource utilization was analyzed in the context of age, gender, race, year, payer status, median household income by zip code quartile (MIQ), hospital bed size, hospital location/teaching status, hospital region, children's hospital status, type of CLM, and type of surgical procedure.

Independent predictors of longer LOS were found using an ordinal logistic regression model (Nagelkerke R^2: 0.688; df: 45). LOS decreased over the study period ($p < 0.001$). Older children had longer LOS with each increasing year in age ($p < 0.001$). Medicaid patients had longer LOS (odds ratio, OR [95% confidence interval, CI] 1.66 [1.18, 2.35], $p = 0.004$) vs. privately insured patients. Medium-sized hospitals had a longer LOS (OR 1.89 [1.19, 2.99], $p = 0.007$) vs. large facilities; along with those located in the Midwestern and Southern U.S. (OR 3.63 [2.22, 6.02] and OR 3.25 [2.21, 4.98] respectively, $p < 0.001$ for both) compared to Western U.S., children's unit in general hospital (CUGH) had longer LOS compared to non-children's hospital (NCH) and children's general hospital (CGH) (OR 0.60 [0.36, 0.99], $p = 0.045$ and OR 0.52 [0.32, 0.84], $p = 0.007$ respectively). Children with CPAM had longer LOS (OR 2.62 [1.31, 5.23], $p = 0.006$) compared to those with a hybrid diagnosis; however LOS for the PS group (11.6 ± 26.5 days) was significantly longer compared to the CPAM group (8.8 ± 14.1 days), $p < 0.001$.

Determinants of TC (R^2: 0.669; df: 44) were identified using a similar method. TC increased significantly over the study period ($p < 0.001$). Older children had higher TC ($p = 0.001$). Medicaid patients had higher TC (OR 1.51 [1.08, 2.11], $p = 0.015$) vs. privately insured patients. The Western U.S. had the highest TC (Northeast OR 0.64 [0.44, 0.93], $p = 0.019$, Midwest OR 0.14 [0.08, 0.22], $p < 0.001$, and South OR 0.17 [0.12, 0.26], $p < 0.001$ vs. West). Children undergoing segmentectomy (OR 2.13 [1.29, 3.53], $p = 0.003$) and lobectomy (OR 1.71 [1.07, 2.73], $p = 0.024$) had higher TC, compared to patients not undergoing segmentectomy or lobectomy, respectively. For a graphical representation of determinants of LOS and TC, see Fig. 1.

2.2. Survival analyses

Cases of CPAM had an in-hospital mortality rate of 1.4%. On bivariate analysis, in-hospital survival was associated with payer status, MIQ, and hospital bed size. Self-pay patients had higher mortality rates (mortality OR 14.6 [2.33, 92.1], $p = 0.039$) vs. privately insured, along with those from households in the lowest MIQ (OR 10.4 [1.25, 87.4], $p = 0.024$). Children treated in small hospitals had lower in-hospital survival rates (mortality OR 7.17 [1.30, 39.6], $p = 0.049$) vs. large facilities.