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Original Article

Laryngotracheobronchial anomalies in infants and the related risk factors of in-hospital mortality

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Abstract

Background: Laryngotracheobronchial anomalies (LTBAs) may cause respiratory problems during early childhood, and increase the risk of hospitalization or mortality in diseased children. This study investigated the initial hospitalization age and risk factors for in-hospital mortality in infants diagnosed with LTBAs during their first 5 years of life.
Methods: Hospitalized infants diagnosed with LTBAs were retrieved from Taiwan's National Health Insurance Research Database from 2003 to 2005. Their medical claim data were traced up to 59 months of age. The age distribution of all LTBA cases was analyzed, and then the enrolled infants were grouped into two age groups. Hospitalization-related comorbidities and risk factors for in-hospital mortality were also analyzed.
Results: A total of 1272 LTBA cases were retrieved. Most of them (976, 76.7%) were initially hospitalized at an age of 0−3 months, and 47 infants (3.7%) died. These enrolled cases were grouped into early and late LTBA groups, with ages of 0−3 months and 4−11 months, respectively. Patients in the late LTBA group had significantly more acute airway infections/asthma and neurological diseases, more frequent hospitalizations, longer hospitalization stay, and higher in-hospital mortality than did the early LTBA group ($p < 0.001$). The adjusted odds ratios (aORs) for in-hospital mortality were significantly higher in the children aged 4−11 months [aOR = 2.50, 95% confidence intervals (CI): 1.36−4.60], or having perinatal disease (aOR = 2.00, 95% CI: 1.07−3.73), cardiovascular disease (aOR = 2.45, 95% CI: 1.30−4.60), other congenital anomalies (aOR = 2.42, 95% CI: 1.28−4.60), and neurological diseases (aOR = 2.32, 95% CI: 1.18−4.53).
Conclusion: Most infants with LTBAs were initially diagnosed and hospitalized when they were aged 3 months or younger. The risk factors for in-hospital mortality of the children with LTBAs included being diagnosed and treated at an age of 4 months and older, and the presence of perinatal disease, cardiovascular anomalies, other congenital anomalies, neurological diseases, and an age of 4 months and older.

Keywords: airway anomaly; airway malacia; hospitalization; infant; in-hospital mortality

Conflicts of interest: The authors declare that they have no conflicts of interest related to the subject matter or materials discussed in this article.

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1. Introduction

Laryngotracheobronchial anomalies (LTBAs) may cause different degrees of respiratory problems which are usually more severe when patients are younger, due to their narrower sizes of airways. These patients may suffer from respiratory distress and airway infections, and require frequent hospitalizations, especially in severe cases. The life quality and growth of the diseased children can be influenced, and the family's burden can be increased. Sometimes, it is a challenge for pediatricians to take care of infants with severe LTBAs. Since LTBAs are not rare among children with pulmonary diseases, an investigation of their hospitalization features and risk factors of mortality during early childhood is important for future care.

LTBAs are heterogeneous diseases including absence, atresia, stenosis and diverticulum of the airway, cartilage anomalies, congenital cleft thyroid and cartilage, laryngocele, and congenital laryngeal stridor.[1−3] Vocal cord paralysis, congenital anomalies of large vessels and abnormalities of the central nervous system may present similar symptoms, and should be taken into differential diagnosis. In mild forms of LTBAs, the symptoms are usually mild and self-limiting by the age of 2 years. Hospitalization is required for patients with comorbidities and other medical conditions including airway infections, other congenital anomalies, gastroesophageal or laryngopharyngeal reflux, and central nervous system diseases.[4−11] Although most patients are diagnosed during early months of life, some are diagnosed during later infancy or childhood.[6,12,13] Different clinical presentations and disease courses between those diagnosed at early or late infancy periods have been investigated.[11] Most reported studies on children's LTBAs have focused on airway malacia, the most common type of LTBAs, and most of them were conducted at a single hospital and only followed up the patients for a short period of time.[2,4,5,8,11,12,14] Thus, an investigation of the clinical outcomes of hospitalized children with LTBAs for multicenter cases and observation for a longer period may provide more information for pediatricians caring for these patients in the future.

The single-payer National Health Insurance (NHI) program in Taiwan was launched in 1995 and covered > 99.5% of the nation's inhabitants.[15] The claims data from Taiwan's National Health Insurance Research Database (NHIRD) have provided trustworthy information for population-based research in children and adults for > 10 years.[16−21] Therefore, it is worthwhile evaluating the clinical outcomes of children with LTBAs using Taiwan's NHIRD for a nationwide, multicentered investigation.

We hypothesized that the diagnosed ages and the presence of comorbidities in infants with LTBAs may influence their clinical outcomes. Therefore, the purpose of this study was to investigate the diagnostic ages and risk factors for in-hospital mortality in hospitalized infants diagnosed with LTBAs during their first 5 years of life.

2. Methods

This study was approved by the Institutional Review Board of Taipei Veterans General Hospital, Taipei, Taiwan (VGH IRB No.2012-06-006A). There was no patient consent required because the data were analyzed anonymously and no personal information could be connected to the data in this study.

2.1. Study population

Hospitalization claims data of Taiwan's NHIRD were used for analysis. The hospitalization claims data contain scrambled and encrypted personal identification numbers, dates of birth, gender, diagnostic codes using the International Classification of Disease, Ninth Revision, Clinical Modification (ICD-9-CM),[22] procedural codes, and admission and discharge dates.

Infants born between 2003 and 2005 and admitted at an age of 0−11 months with a discharge diagnosis of LTBAs (ICD-9-CM code: 748.3) were retrieved for analysis. The hospitalization records of all patients were traced until they were 59 months of age. The enrolled infants were grouped into two age groups according to the findings of age distribution and compared.

Data on hospitalization frequency, total hospital stay, age at receiving the earliest bronchoscopy examinations (ICD-9-CM codes: 31.72, 33.22, 33.23, 33.24, 33.26, 33.27, and 31.72) and tracheostomy (ICD-9-CM codes: 31.1, 31.21, and 31.29), and the discharge diagnoses and status were retrieved and analyzed. Clinical Classification Software (CCS) 2010 version was used to classify the diagnoses into clinically meaningful categories.[23] Acute airway infections included the diagnoses of pneumonia, bronchitis, and other upper and lower airway infections (CCS codes: 122, 125, 133, 134).[23]Special attention was also paid to the categories of cardiovascular anomalies (CCS code: 213), other congenital anomalies (CCS code: 217), perinatal diseases (CCS code: 224), esophageal diseases (CCS code: 138), and neurological diseases (including epilepsy and other central nervous system disease (CCS code: 83) for which the discharge diagnoses were further classified and analyzed by the first three digits of the ICD-9-CM codes.[22,23]

2.2. Data analysis

The database management software PostgreSQL (version 9.34, The PostgreSQL Global Development Group, Los Angeles, CA, USA) was used for data processing,[24] and SPSS 22.0 (IBM Corp., Armonk, NY, USA) was used for statistical analysis. The Mann-Whitney *U* test was used for comparisons between two groups with continuous variables, and the Chi-square test was used to compare categorical variables. Logistic regression modeling was used to analyze the odds ratio (ORs) for associated factors with the in-hospital mortality rate among the patients with LTBAs. The decision threshold for eligibility was $p < 0.10$ for the univariate model and to remain in the multivariate model. A p value < 0.05 was used to determine statistical significance.

3. Results

From the nationwide claims of 11,642,140 hospitalizations, there were 173,785 (1.5%) infants aged 0−11 months who