THE EPIDEMIOLOGY OF LARYNGEAL STENOSIS IN INFANTS: A POPULATION-BASED APPROACH
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INTRODUCTION
Laryngeal stenosis and more specifically subglottic stenosis has long been recognized as a source of significant morbidity in both adults and children. Arguably the most important advancement regarding management of the neonatal airway was popularized in 1963 by McDonald and Stocks when they presented their experience with prolonged nasotracheal intubation at the Royal Children’s Hospital in Melbourne, Australia. Healthcare utilization for children with laryngeal stenosis has been demonstrated to be significant. Shah et al reported that in 2003, the mean hospital charges for a child admitted with a diagnosis of subglottic stenosis was over $50,000 with a mean length of stay over 11 days. Furthermore, when children greater than 12 months of age were excluded, the mean charges were greater than $85,000 and the mean length of stay increased to over 19 days. Given the impact of individual cases and the overall healthcare burden, it is important to have a marker of the true incidence of this condition in the United States.

Given the current lack of a national understanding of the epidemiology of infants with laryngeal stenosis, the primary goal of the project is to define population-based estimates of the incidence of infants affected with laryngeal stenosis in the United States in a way that is reproducible and can be tracked over time. Secondary goals are to understand factors associated with a laryngeal stenosis diagnosis and determine the characteristics of operative intervention in this patient population.

METHODS AND MATERIALS
This study examined inpatient admissions for pediatric caustic ingestion injuries using the Kids’ Inpatient Database (KID), which is part of the Healthcare Cost and Utilization Project (HCUP) sponsored by the AHRQ. The KID provides data on a sample of all discharges of children aged 20 years and younger in the United States during a given year. The data are obtained from administrative records and reported by hospitals in participating states. These data are subsequently compiled into a public-use dataset.

The KID was accessed for this project. The data are a representative sample of discharges occurring in the United States during 2009 and are well described by the AHRQ. The total number of discharges sampled in 2009 was 3,407,146. The sampling strategy is simple stage and was designed to select 10% of uncomplicated hospital births and 90% of complicated births and other hospital admissions from each state.

Descriptive analysis was performed using Microsoft Excel 2007 (Redmond, WA) and R (R Development Core Team, 2010). The National Vital Statistics Reports are published by the U.S. Department of Health and Human Services through the Centers for Disease Control and Prevention. Live birth and infant mortality data is publicly accessible. The final data set for 2009 was accessed. The availability of specific population level data regarding laryngeal stenosis and infant mortality was included in the analysis of the infant population during the KID 2009 sampling timeframe thus allowing an estimation of the annual incidence of laryngeal stenosis nationally. For the purposes of this report, the term infant refers to any child less than 12 months of age. Statistical Analysis
The primary analysis was to generate descriptive statistics of the dataset. All analyses were performed at the State IC level using 1.22 (Califorina) weights. All national data estimates were calculated using sampling weights based on the database sampling scheme. As the data represent sample-based estimates, 95% confidence intervals are presented. Regression analyses were performed at the State IC sampling environment.

Logistic regression models were constructed to identify potential factors related to the dependent outcome of a diagnosis of laryngeal stenosis as well as the outcomes of underlying airway-related surgical procedures in the setting of laryngeal stenosis. Procedural outcomes included tracheostomy placement alone, endoscopic or open airway intervention without tracheostomy placement or any therapeutic intervention procedure inclusive of both primary and secondary interventions. Independent variables included patient factors as well as hospital factors. Patient factors selected for analysis were gender, race, and median income quartile of the patient’s ZIP code (selected as a proxy of economic status). Hospital factors included region within the United States (Northeast, Midwest, South, West), hospital location (rural vs. urban), and hospital teaching status (non-teaching vs. teaching). Statistical significance was set at p<0.05.

RESULTS
Summary Estimates
The age-adjusted prevalence estimates of hospitalized infants less than 12 months of age diagnosed with laryngeal stenosis in 2009 are presented in Table 1. The prevalence in this population is estimated to be 1374 (95%CI 1313, 1415) infants with a male predominance. Of the infants diagnosed with laryngeal stenosis, 28% underwent at least one operative airway intervention including dilatation, endoscopic or open/grafting, and tracheostomy placement indicating increased severity.

Figure 1 demonstrates the race of infants diagnosed with laryngeal stenosis during this time period. Figure 2 demonstrates the median income quartile for the zip code listed on admission. Reporting hospital characteristics, 9.8% (95% CI 8.5%, 11.2%) of the diagnoses were made in teaching hospitals and 98.2% (95% CI 97.6%, 98.7%). Regional variation is demonstrated in Figure 3.

Incidence Estimates
The CDC reports that there were 4,130,665 live births in 2009 as well as 26,408 infant deaths. Therefore, the national live population was calculated to be 4,104,237.

Based on this data, the annual incidence of laryngeal stenosis can be estimated to be 33.5 (95%CI 27.6, 39.3) cases per 100,000 infants. When analyzing based on severity requiring an operative airway intervention, the annual incidence decreases to an estimate of 7.5 (95%CI 3.9, 12.1) cases per 100,000 infants. The annual incidence of laryngeal stenosis requiring a tracheostomy is estimated to be 4.0 (95%CI 2.8, 5.3) cases per 100,000 infants. Regression Analysis
Table 2 demonstrates the results of the logistic regression analysis designed to identify potential factors associated with the diagnosis of laryngeal stenosis in infants. The model includes an association between racial stenosis and race as a member of the African American race (p<0.0001). Hospital characteristics include identification as a teaching hospital (p<0.0001) in an urban location (p<0.046) that is not located in the southern region of the United States (p<0.0001). Of note, elective compared to urgent/emergent admissions are associated with an increased odds of being diagnosed with laryngeal stenosis (p<0.0001).

Logistic regression analyses regarding procedural data include infants undergoing a tracheostomy alone (Table 3), an open or endoscopic airway intervention without a tracheostomy (Table 4) or undergoing any therapeutic procedural intervention of tracheostomy (Table 5). Of note, infants undergoing tracheostomy alone were associated with a median admission (or were never discharged prior to receiving a tracheostomy) while children undergoing airway interventions without tracheostomy placement were associated with elective admissions. Aside from a slight association with males receiving more tracheostomies alone, there were no gender, racial or income quartile differences within the various procedure categories.

CONCLUSIONS
Despite well-documented successes in prevention, the true incidence of laryngeal stenosis in infants is likely higher than previously indicated, but of a variable severity. The inability of population level to characterize this condition provides a crucial means to understand and analyze the associated epidemiologic factors as well as the ability to track this relatively rare disease given the known public health burden.

REFERENCES