Analysis of Vocal Fold Motion Impairment in Neonates Undergoing Congenital Heart Surgery

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IMPORTANCE Vocal fold motion impairment (VFMI) is a known risk factor following congenital heart surgery (CHS). The impact of this diagnosis on utilization and outcomes is unknown.

OBJECTIVE To evaluate the cost, postprocedure length of stay (PPLOS), and outcomes for neonates with VFMI after CHS.

DESIGN, SETTING, AND PARTICIPANTS A cross-sectional analysis of the 2012 Kids' Inpatient Database (KID) of neonates who underwent CHS was carried out. The KID is an administrative data set of patients, aged 20 years or younger, and contains data on more than 10 million hospitalizations from 44 states. The KID is limited to inpatient hospitalization and contains discharge summary level of data. Patients were limited to those who were born during the hospitalization and those who were aged 28 days or younger at the time of admission for CHS. A weighted total of 4139 neonates who underwent CHS were identified, of which 3725 survived. The proportion of neonates diagnosed with VFMI was 264 (6.92%) of 3725.

EXPOSURES Congenital heart surgery.

MAIN OUTCOMES AND MEASURES Cost of inpatient hospital stay, postprocedure length of stay, odds of pneumonia, gastrostomy tube placement, and tracheostomy tube placement. Risk-adjusted generalized linear models examined differences in cost and PPLOS between neonates who underwent CHS and were diagnosed with VFMI and those who were not. Risk-adjusted logistic regression compared the odds of selected outcomes (gastrostomy, tracheostomy, pneumonia). Models were weighted to provide national estimates.

RESULTS Of 3725 neonates (aged 0-28 days), 2203 (59.1%) were male and 1517 (40.7%) were female. Neonates diagnosed with VFMI had significantly higher total cost by $34 000 (95% CI, 2200-65 000) and PPLOS by 9.1 days (95% CI, 4.6-13.7) compared with those who did not. When PPLOS was included as a covariate in the model for cost, presence of VFMI was no longer significant. There were no differences in odds of pneumonia, gastrostomy, or tracheostomy.

CONCLUSIONS AND RELEVANCE Vocal fold motion impairment after CHS was associated with significant increases in cost owing to increased PPLOS. These findings provide a foundation to further investigate standardized screening for VFMI following CHS; early identification and treatment may decrease cost and PPLOS.

Published online March 15, 2018.

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Pediatric voice and swallowing disorders affect an estimated 1% to 9% of children in the United States, which translates to up to approximately 6 million children affected by these problems. More than 500,000 children in the United States are diagnosed with dysphagia annually. However, given inaccurate parental reports, and less than 25% of parents seeking medical treatment, the true burden of voice and swallowing disorders are likely underestimated. The downstream effects of dysphagia and dysphonia can be considerable, including increased risk for aspiration-induced lung injury, sequelae associated with malnutrition, and caregiver stress and anxiety over social stigma of voice abnormalities.

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Pediatric vocal fold motion impairment (VFMI) is a well-known cause of dysphonia and dysphagia. The association of VFMI with prior cardiothoracic surgery has been well-established by a number of studies and characterization of the anatomic relationship of the RLN and cardiothoracic structures demonstrates causation. In particular, the course of the left RLN places it at increased risk, with its longer intrathoracic course, descending medially past the patent ductus arteriosus (PDA), looping around the aortic arch, then ascending medially toward the tracheoesophageal groove. Proposed mechanisms of injury include RLN compression by a cuffed endotracheal tube, injury to the cricothyroid or cricoarytenoid joints, thermal trauma, use of intraoperative transesophageal echocardiography with RLN compression at the TE groove, median sternotomy with resultant bilateral RLN strain, and direct injury to the nerve by transection or stretching, particularly in procedures involving manipulation of the aortic arch. In addition, low birth weight and prolonged intubation have been associated with VFMI following congenital heart surgery (CHS).

Multiple other investigators have attempted to characterize the incidence of VFMI following cardiothoracic surgery, but the lack of a uniform protocol and the retrospective nature of many of these studies have made them subject to selection bias, with possible under or overestimation of the incidence of VFMI. The prospective studies have looked primarily at PDA ligation or have looked at all pediatric cardiac surgery, without specifying a particular age group or type of surgery. Limited studies on long-term outcomes in patients with VFMI after CHS suggests an increase in hospital admissions related to feeding and respiratory complications. However, overall, there is little on the subject of cost and resource utilization for this group of patients.

To begin to quantify the effect of VFMI after CHS on patient outcomes and the health care burden, we sought to answer the following questions using the Kids’ Inpatient Database (KID): first, is VFMI after CHS associated with an increase in the cost and postprocedure length of stay (PPLOS) of hospitalization for surgery? And second, is there a difference in outcomes, including pneumonia, tracheostomy tube placement, and gastrostomy tube placement, during hospitalization for cardiac surgery if a diagnosis of VFMI is made?

Key Points
Question Do differences in cost, postprocedure length of stay (PPLOS), and outcomes for neonates with and without vocal fold motion impairment (VFMI) after congenital heart surgery exist?
Findings In this cross-sectional analysis of 3725 neonates, the proportion diagnosed with VFMI after congenital heart surgery was 6.9%. Neonates diagnosed with VFMI had significantly higher total hospital cost and PPLOS compared with those who did not; there were no differences in odds of pneumonia, gastrostomy, or tracheostomy.
Meaning Vocal fold motion impairment following congenital heart surgery was associated with increased cost and PPLOS, and protocols for early identification of VFMI or techniques to prevent VFMI may result in a decrease in cost and PPLOS.

Methods
Data on patient encounters were analyzed from the Agency for Healthcare Research and Quality (AHRQ)–sponsored Healthcare Cost and Utilization Project (HCUP) KID from 2012. The KID is an administrative data set of patients, aged 20 years or younger, and contains data on more than 10 million hospitalizations from 44 states. The KID uses a sampling of pediatric and hospitalization and contains discharge summary level of data.

Patients who were admitted in the neonatal period and underwent CHS during the same hospitalization were identified in the KID using the International Classification of Diseases, Ninth Revision, Clinical Modification (ICD-9-CM) principal procedures codes for congenital heart surgery (Supplement). Patients were limited to those who were born during the hospitalization and those who were aged 28 days or younger at the time of admission for CHS. This was done to account for those who were born in and underwent CHS at the same hospital, as well as those who were transferred to another hospital during the neonatal period for CHS. Outcomes were grouped relative to the presence or absence of VFMI. The Emory University Institutional Review Board deemed this study exempt from ethical review because all data were deidentified.

Outcome Measures
The primary outcome measures examined differences in total hospital cost and PPLOS between neonates who underwent CHS and were diagnosed with unilateral or bilateral VFMI (ICD-9-CM diagnosis codes 344.9, 478.30, 478.31, 478.32, 478.33, and 478.34) and those who were not. Hospital costs were calculated from total hospital charges by multiplying charges by hospital-specific cost-to-charge ratios (CCRs, created by dividing the inpatient costs by the inpatient charges; does not
include professional fees) for the corresponding year. Costs were converted to 2012 US dollars using annual Hospital Consumer Price Indices. The PPLOS was calculated by subtracting the day of surgery from the total LOS. Secondary outcomes included selected perioperative complications that may affect PPLOS and cost.

### Statistical Analyses

Statistical analyses were performed using Stata statistical software (version 13.1, StataCorp LP). All tests of significance were 2-sided ($P < .05$). Generalized linear models (family $\gamma$, link log) followed by postestimation of average marginal effects were used to attain risk-adjusted predicted mean differences and standard errors for nonnormally distributed continuous total hospital cost and PPLOS. Multivariable logistic regression was used to attain risk-adjusted odds ratios (ORs) and corresponding 95% confidence intervals (95% CIs) for the binary outcome of complication. All models were risk-adjusted, using included covariates. Multilevel models with robust standard errors accounted for clustering of patients in hospitals. Models were further weighted using KID-provided design weights to attain nationally representative effects.

### Results

#### Patient Characteristics

An unweighted total of 2913 admissions for CHS were included. The weighted national sample consisted of 4139 patients, of which 3725 survived. A weighted total of 264 patients (6.92%) with VFMI after CHS were identified. In patients who underwent a Norwood procedure, a weighted proportion of 50 of 422 (11.9%) with VFMI after surgery were identified. Of 3725 patients, 265 (7.1%) patients who underwent CHS had flexible fiberoptic laryngoscopy or direct laryngoscopy performed and 1088 (29%) who underwent CHS had a diagnosis of feeding difficulty/dysphagia.

Table 1 presents differences in patient demographic characteristics, comparing those with VFMI after CHS with those without. Of note, those who developed VFMI after CHS were significantly more likely to have Medicaid/Medicare insurance, have had surgery in the Midwest, and have a significantly higher RACHS-1 on average. Of note, when patients with a RACHS-1 score of 6 (Norwood procedure) were excluded, the groups are comparable (RACHS-1 in patients with VFMI was 3.42; 95% CI, 3.29-3.55; RACHS-1 in non-VFMI patients was 3.28; 95% CI, 3.24-3.31).

#### Unadjusted Cost and Outcomes Results

Unadjusted total hospital costs, PPLOS, and complications are presented in Table 2. Weighted differences in 2012 US dollars reveals costs were higher for patients who developed VFMI after CHS, with total median hospital costs of nearly $171,000 (IQR, $131,000-$291,000). Total hospital costs for those who did not develop VFMI were approximately $127,000 (IQR, $82,000-$211,000). Unadjusted median PPLOS for VFMI vs no VFMI was 34 days (IQR, 20-50 days) vs 18 days (IQR, 10-34 days), ranging from 1 to 6 to each patient based on type of surgery, age, and comparable in-hospital mortality. Further details regarding generation of this score can be found in the cited work by Jenkins et al.\textsuperscript{29} Patients with a diagnosis of necrotizing enterocolitis (NEC) and feeding difficulty/dysphagia were also identified because these were thought to be additional complications that may affect PPLOS and cost.
respective. Complications of interest, including pneumonia, tracheostomy, and gastrostomy tube placement, were low for both groups.

**Multivariable Cost and Outcomes Results**

Risk-adjusted total hospital cost and outcomes are presented in Table 3 and Table 4. Mean adjusted cost increase associated with VFMI was $34,123 (95% CI, $3636-$64,610) per patient admission. Other covariates that affected cost are as shown in Table 3, including a diagnosis of NEC or pneumonia during the hospital course. Of note, when Norwood patients were excluded from the analysis, the mean adjusted cost increase associated with VFMI increased to $37,305 (95% CI, $5570-$69,039). When only patients with unilateral VFMI were included, this effect persisted, with a cost difference of $35,391 (95% CI, $39,395-$66,847). When only the Norwood group was analyzed, no difference in cost was seen.

In addition, the mean PPLOS increase was 9.1 days (95% CI, 4.6-13.7 days). Other covariates that affected PPLOS are as shown in Table 3. When Norwood patients were excluded from the analysis, the increase in PPLOS associated with VFMI increased to 10.5 days (95% CI, 5.6-15.4 days). When only patients with unilateral VFMI were included, this effect persisted, with a PPLOS difference of 9.6 days (95% CI, 4.8-14.4 days). When only the Norwood group was analyzed, no difference in PPLOS was seen.

There were no significant differences in odds of pneumonia, tracheostomy, or gastrostomy tube placement between the 2 groups. Counts for all of these outcomes were low for both groups.

**Discussion**

Though VFMI is a known complication of CHS, there is limited literature on its impact on the health care system or on patient outcomes. To our knowledge, this is the first population-based study to examine inpatient outcomes for VFMI following CHS. These selected outcomes were chosen because they are potential complications/procedures for which vocal fold motion impairment has been found to be a risk factor.²⁰,³⁰-³² We found that neonates who are diagnosed with VFMI during the inpatient hospitalization had an associated increase in total hospital costs that could be attributed in increased PPLOS; however, there was no difference in rates of pneumonia, gastrostomy tube placement, or tracheostomy. We do suspect that there may have been inaccurate coding for these outcomes, given overall low counts.

In our study, 264 (6.9%) of the 3725 surviving neonates who underwent CHS had a documented diagnosis of VFMI during the inpatient hospitalization. This is consistent with the estimated incidence in the literature.¹⁹,²¹-²⁵,³¹,³³-⁴⁵ This estimate of incidence faces the same bias noted in other retrospective studies, namely that a selection bias may underestimate the incidence because not all patients are screened for VFMI after CHS. Given this variability in the established incidence, development of a screening protocol based on specific risk criteria is a crucial next step in treating these patients.

**Table 2. Unadjusted Costs and Outcomes Results**

<table>
<thead>
<tr>
<th>Outcome</th>
<th>VFMI After CHS (n = 264)</th>
<th>No VFMI After CHS (n = 3461)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total hospital costs in dollars, median (IQR)</td>
<td>170,671 (130,835-291,552)</td>
<td>127,468 (82,134-210,590)</td>
</tr>
<tr>
<td>Postprocedure length of stay in days, median (IQR)</td>
<td>34 (20-50)</td>
<td>18 (10-34)</td>
</tr>
<tr>
<td>Complications, No. (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pneumonia</td>
<td>&lt;10</td>
<td>57 (1.47)</td>
</tr>
<tr>
<td>Tracheostomy</td>
<td>&lt;10</td>
<td>75 (1.94)</td>
</tr>
<tr>
<td>Gastrostomy tube placement</td>
<td>&lt;10</td>
<td>&lt;10</td>
</tr>
</tbody>
</table>

Abbreviations: CHS, congenital heart surgery; VFMI, vocal fold motion impairment.

The significance of determining true incidence of this entity should not be overlooked because underestimation can result in wasted resources from delayed identification and a loss of appropriate health care and research funds to help treat these patients. Routine screening would increase the identification of asymptomatic cases and would also help identify whether respiratory and feeding difficulties are associated with VFMI, or whether these symptoms are attributable to the underlying cardiac disease and postsurgical recovery. When feeding difficulties are substituted for VFMI in the model, there is also an increased PPLOS. We believe that if more patients were screened for VFMI, particularly those with feeding difficulty, changes in postoperative feeding management may take place, reducing PPLOS and cost. In addition, accurate knowledge of incidence can help with improved family counseling in the preoperative setting. The data from this study suggest that development of a protocol by which to identify and treat those with VFMI after CHS may result in improved health care resource utilization during the surgical hospital stay.

We hypothesize that the increased PPLOS between the 2 groups is likely related to feeding difficulty because there is no standard protocol on treating these patients from a feeding standpoint. Multiple studies have demonstrated the correlation of VFMI following cardiothoracic surgery with increased length of stay (LOS), dysphagia necessitating alternative forms of nutrition, risk of respiratory disease, and cost of care.¹⁹,²⁰,³³,³⁴,³⁷,⁴¹,⁴² In the 2009 study by Benjamin et al,³⁴ examining extremely low birth weight infants undergoing PDA ligation, VFMI was associated with significantly increased risk of bronchopulmonary dysplasia, reactive airway disease, and need for gastrostomy tube placement. In a series of studies from Texas Children's Hospital, inpatient LOS was noted to be 12 days longer on average for those with VFMI, compared with those without VFMI, and only 50% of those patients went home on a regular diet by mouth, compared with 94% of those without VFMI.¹⁹ In addition, these patients were 7-fold more likely to be admitted for poor weight gain or feeding issues.⁴¹ Our study demonstrates that in the acute inpatient setting, VFMI after CHS is indeed associated with increased PPLOS and therefore increased cost. It is possible that by screening early for VFMI in the appropriate patients, PPLOS can be decreased by formulating a feeding plan, possibly early
injection laryngoplasty, and arranging for outpatient supplies earlier in the hospital course.

As survival of patients with congenital heart disease has improved significantly since the introduction of surgical palliation and cardiac transplantation, the disease has morphed from a lethal disease into a chronic one.\textsuperscript{46-48} Therefore, there has been a shift away from solely focusing on mortality to also looking at other aspects of care. Elucidating short- and long-term outcomes, including PPLOS, time to diagnosis of VFMI, time to initiation of oral feeding, respiratory and feeding difficulties and poor weight gain for neonates with VFMI after CHS is a critical step to improving outcomes for this population. Analysis of these data will lead to the development of benchmark values on which improvements can be made with the advent of new protocols for screening. Earlier diagnosis will lead to improved short- and long-term outcomes, including PPLOS, decrease time to oral feeding, earlier otolaryngologic intervention if indicated, and decreased rates of readmission for pulmonary or feeding complications.

**Strengths and Limitations**

Strengths of this study include a large sample size and professional coding used by the Agency for Healthcare Research and Quality in the KID. Administrative databases have the ability to support a well-powered study with statistically valid data, and use weights to provide a nationally representative sample. Large confidence intervals for the primary outcome of interest for both cost and PPLOS do exist, and we believe the large confidence intervals are likely owing to factors such as variability in hospital practices or other patient comorbidities. Given the large sample size and the degree of significance of the findings, we do feel the results accurately represent important differences in cost and PPLOS seen between the 2 groups. There are, however, several covariates that are associated with PPLOS and cost. This warrants further investigation of whether there is any other data that may explain this, such as difference in surgical techniques or postprocedure feeding protocols. For instance, we suspect that there is a difference in which hospitals prefer to send patients home with a nasogastric tube vs gastrostomy tube. In addition, there may be social barriers leading to less timely discharge in patients of particular races.

There are several limitations to this analysis. The use of an administrative database is limited by lack of potentially important patient-level clinical data that may explain some of these observed differences between the groups. We cannot identify at which point during the hospitalization the VFMI was identified and whether early identification improves outcomes. Selection bias may underestimate the true incidence because not all patients are screened for VFMI after CHS.

### Table 3. Risk-Adjusted Cost Regression Results, Weighted Data\textsuperscript{*}

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean Difference From Baseline (95%CI)</th>
<th>Postprocedure Length of Stay, d</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Total Hospital Cost (2012 Dollars)</td>
<td></td>
</tr>
<tr>
<td>No VFMI</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>VFMI\textsuperscript{*}</td>
<td>33 614 (2170 to 64 609)</td>
<td>9.1 (4.6 to 13.7)</td>
</tr>
<tr>
<td>Male</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>Female</td>
<td>18 972 (4448 to 33 495)</td>
<td>4.2 (1.2 to 7.1)</td>
</tr>
<tr>
<td>Non-Hispanic white</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>Non-Hispanic black</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>First income quartile</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>Second income quartile</td>
<td>$25 145 (−46 493 to −3798)</td>
<td>−3.9 (−8.5 to 0.8)</td>
</tr>
<tr>
<td>Third income quartile</td>
<td>$19 139 (−38 269 to −9)</td>
<td>−4.3 (−7.9 to −0.7)</td>
</tr>
<tr>
<td>4th income quartile</td>
<td>$11 300 (−34 201 to 11 690)</td>
<td>−6.7 (−10.1 to −3.2)</td>
</tr>
<tr>
<td>Northeast</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>Midwest</td>
<td>65 006 (20 757 to 109 254)</td>
<td>5.9 (0.9 to 11.0)</td>
</tr>
<tr>
<td>South</td>
<td>48 106 (10 665 to 85 547)</td>
<td>8.1 (4.1 to 12.1)</td>
</tr>
<tr>
<td>West</td>
<td>53 087 (17 275 to 88 899)</td>
<td>NS</td>
</tr>
<tr>
<td>No NEC</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>NEC</td>
<td>116 169 (67 390 to 165 028)</td>
<td>18.5 (11.3 to 25.7)</td>
</tr>
<tr>
<td>No pneumonia</td>
<td>0 [Reference]</td>
<td>0 [Reference]</td>
</tr>
<tr>
<td>Pneumonia</td>
<td>172 987 (122 005 to 223 969)</td>
<td>31.1 (21.1 to 39.1)</td>
</tr>
</tbody>
</table>

**Abbreviations:** CHS, congenital heart surgery; NS, not significant; VFMI, vocal fold motion impairment.

\textsuperscript{*} Mean difference: for example, average total hospital costs for patients with VFMI after CHS were about $34 000 (95% CI, \$2200-$65 000) higher than average total hospital costs for patients without VFMI after CHS.

### Table 4. Risk-Adjusted Outcome Regression Results, Weighted Data\textsuperscript{*}

<table>
<thead>
<tr>
<th>Variable</th>
<th>Odds Ratio (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pneumonia</td>
</tr>
<tr>
<td>No VFMI</td>
<td>1 [Reference]</td>
</tr>
<tr>
<td>VFMI\textsuperscript{*}</td>
<td>0.57 (0.13-2.40)</td>
</tr>
</tbody>
</table>

**Abbreviation:** VFMI, vocal fold motion impairment.

\textsuperscript{*} Odds ratio: the odds of pneumonia in patients with VFMI after CHS were 0.63-times (95% CI, 0.16-2.48) the odds of pneumonia in patients without VFMI after CHS.
There were also limitations when attempting to evaluate impact of bilateral or unilateral vocal fold paralysis and possible interventions these patients may have received because the number of patients with bilateral VFMI and the number of neonates with VFMI after CHS who had inpatient injection laryngoplasty were both so small that they cannot be reported. The database also did not contain many instances where speech language pathology rehabilitation services were used based on codes. The KID is a cross-sectional database and cannot provide longitudinal data on outpatient visits or readmissions. Just as long-term outcomes cannot be assessed with this database, clinically relevant measures, such as quality of life, long-term feeding, and speech outcomes cannot be reported.

The next steps in identifying factors that may improve outcomes in this population include a prospective study to evaluate clinical aspects of this patient population and development of a longitudinal database to identify cost, resource utilization, and outcomes over time. This information can be used toward the development of a targeted protocol for screening of patients who are more likely to have VFMI based on patient and surgery characteristics. The goal is to evaluate if early screening improves PPLOS and feeding outcomes. Early screening may lead to considering early injection that may alleviate the need for full nasogastric tube feeds. It will also allow speech pathologists to more deliberately plan follow-up, especially when further radiographic studies are indicated based on the expected clinical course. We offer our findings to contribute to an evidence-based practice guideline that will decrease practice variation. By doing so, we can decrease unnecessary resource utilization and uniformly improve outcomes.

Conclusions

Vocal fold motion impairment after CHS is a considerable risk factor and results in increases in cost owing to increased PPLOS. These findings provide a foundation to further investigate universal and/or early screening following particular CHS. Screening protocols should be established to improve outcomes and decrease resource utilization and costs associated with this complication.

REFERENCES


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