Outcomes of Vocal Fold Motion Impairment and Dysphagia after Pediatric Cardiothoracic Surgery: A Systematic Review

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Abstract

Objective. The objective of this study was to systematically review the literature regarding vocal fold motion impairment (VFMI), respiratory outcomes, and swallowing outcomes in children following congenital heart surgery (CHS).

Data Sources. PubMed, Embase, Medline, and CINAHL databases.

Review Methods. Data sources were searched from inception to November 30, 2018. Studies that described recovery of VFMI and swallowing function following CHS were included, and a qualitative analysis was performed.

Results. A total of 1371 studies were identified, of which 8 met inclusion criteria for VFMI and 5 met inclusion criteria for swallowing outcomes. Studies including patients who underwent isolated patent ductus arteriosus ligation were excluded. VFMI was present in 8% to 59% of subjects, and rates of recovery ranged from 9% to 96% at 6 months to 6 years of follow-up. Inability to maintain an oral diet occurred in 14% to 100% of subjects with VFMI and 11% to 61% without VFMI following surgery. Tolerance of an oral diet without tube feeding was present in 66% to 75% of subjects with VFMI and 88% to 100% without VFMI at 24 days to 3.2 years of follow-up. Limited data suggest that time to extubation is longer in VFMI subjects, but overall hospital length of stay and mortality may not be affected by VFMI status.

Conclusions. Data evaluating dysphagia and VFMI after CHS are limited. Most studies suggest significant improvement in swallowing function, while rate of recovery of VFMI is variable. Future prospective studies with standardized screening and follow-up are needed to better elucidate outcomes to help develop algorithms for identification and management of VFMI after CHS.

Keywords

pediatric, congenital heart surgery, vocal fold motion impairment, dysphonia, dysphagia, recovery

Received January 30, 2019; accepted May 31, 2019.

Congenital cardiac defects are common, affecting approximately 40,000 births per year.1,2 Approximately one-third of these patients will have a severe defect and require surgical repair,3,4 which may involve open heart surgery or transcatheter repair. The recurrent laryngeal nerves (RLNs) are at risk for iatrogenic injury during cardiac surgery, particularly if a sternotomy is performed.5

RLN injury can result in vocal fold motion impairment (VFMI), which may lead to significant voice, respiratory, and feeding difficulties. Respiratory symptoms of VFMI are typically more prominent with bilateral VFMI and include difficulty breathing, stridor, and, in severe cases, respiratory failure requiring mechanical ventilation and/or tracheotomy. Feeding difficulties include poor feeding, malnutrition, aspiration, increased work of breathing with feeding, failure to thrive, and need for nasogastric or gastric tube feedings. Laryngopharyngeal dysfunction is recognized as one of the more common morbidities associated with surgery for congenital cardiac conditions.6,7 Accordingly, the literature supports routine postoperative evaluation of vocal fold motion.8

Outcomes data for VFMI and swallowing dysfunction following congenital heart surgery (CHS) are difficult to

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characterize in the current literature. While there are many studies examining the incidence of VFMI and dysphagia after surgery, there are relatively fewer studies on the outcomes and progression related to RLN injury. Many studies suggest that recovery or significant improvement regarding VFMI \(^{9,10}\) and swallowing \(^{11}\) is expected in a significant proportion of subjects within several months after surgery, whereas others did not demonstrate significant recovery of either in their cohorts. \(^{12,13}\) Furthermore, the individual studies often have slightly different objectives and inclusion criteria, thereby complicating critical analysis of the literature. This leaves clinicians in the precarious position of making decisions without the support of evidence-based medicine and being unable to provide families with a prognosis regarding their children’s VFMI and feeding capabilities. This systematic review aims to answer the following questions: (1) For children undergoing CHS, does postoperative VFMI as compared with normal vocal fold motion result in worse swallowing and respiratory outcomes in the short and long term? (2) For children undergoing CHS who develop VMFI, what is the rate of recovery of vocal fold motion?

**Methods**

**Guidelines**

Though no randomized clinical trials were identified for inclusion in this systematic review, the PRISMA recommendations (Preferred Reporting Items for Systematic Reviews and Meta-analyses) were used.\(^{14,15}\)

**Information Sources**

PubMed, EMBASE, Medline, and CINAHL databases were queried from inception until November 30, 2018. The search was performed by a medical librarian and included all studies limited to English language and human subjects.

**Search**

The following MeSH terms (Medical Subject Headings) were first used to identify articles of potential interest: (((vocal fold OR vocal cord) AND (dysphagia OR aspirate OR aspiration OR feed OR feeding OR swallow OR swallowing OR dysphonia OR hoarseness OR hoarse OR paralysis OR palsy OR paresis OR dysfunction OR immobility OR immobilization OR immobilisation OR hypomobility)) OR “laryngeal paralysis” OR “laryngeal palsy” OR “laryngeal paresis” OR “laryngeal dysfunction” OR “laryngeal immobility” OR “laryngeal immobilization” OR “laryngeal immobilisation” OR “laryngeal hypomobility”) AND (cardiac OR cardiovascular OR cardiothoracic OR aorta OR aortic OR heart OR ductus arteriosus OR PDA OR norwood OR hypoplastic OR HLHS or congenital) AND (neonatal OR neonate OR child OR infant OR newborn OR pediatric OR pediatric). Articles mapping to the MeSH search terms were used to select the pediatric population. All 3 groups of articles were cross-referenced and limited to human subjects.

**Study Selection**

Articles were excluded if they were not in English or not available in full text, if they were duplicates or nonhuman studies, if they included adult subjects, or if they were single case reports or did not pertain to the outcomes of interest. Additionally, any study of subjects who had undergone patent ductus arteriosus (PDA) ligation in isolation was removed due to significant differences between this population and subjects undergoing open cardiac procedures. References of the identified studies were reviewed to identify additional studies.

Five authors (S.O., R.J., J.O., J.B., and N.R.) worked independently to evaluate titles and abstracts to identify articles of interest and to exclude articles that did not meet inclusion criteria. Full articles were obtained for the remaining list of abstracts, and 2 authors (S.O. and R.J.) worked independently to evaluate and determine a final list of studies to be included in the analysis. The lists were then compared, and any conflicts were resolved after further evaluation and discussion regarding the manuscripts.

**Outcomes and Data Extraction**

Data extraction focused on 3 categories of outcomes: VFMI outcomes, respiratory outcomes, and dysphagia outcomes. VFMI outcomes included the incidence of VFMI, laterality of the VMFI, number of subjects who recovered VF motion, and time to VF motion recovery. Respiratory outcomes were defined as the need for ventilator support, need for tracheotomy, number of patients successfully undergoing decannulation, and time to decannulation. Swallowing outcomes included incidence of dysphagia, need for enteral feeding (nasogastric or gastrostomy tubes), and time to initiation of an oral diet. Demographic and other basic data were collected, including average age of the cohort, weight at surgery, type of surgery, mortality, follow-up period, and protocol used for identifying VFMI in each study. Due to heterogeneity of the data found in the identified studies, descriptive statistics were used, with no meta-analysis performed.

**Evaluation of Risk of Bias**

Two authors independently reviewed the articles for risk of any bias using the US National Institutes of Health’s Quality Assessment of Case Series Studies tool,\(^{16}\) and results are reported in **Table 1**. Any discrepancies were resolved by involving a third coauthor to make the final decision.

**Results**

**Literature Selection and Characteristics**

A total of 1371 studies were identified, and 8 met inclusion criteria (**Figure 1**). All 8 were included in the VFMI subset (n = 515), and 5 were included in the dysphagia subset (n = 374). There were 2 retrospective case series and 6 retrospective cohort studies included in this review.
Table 1. Assessment of Risk of Bias.

<table>
<thead>
<tr>
<th>Author</th>
<th>Outcomes Analyzed</th>
<th>Items</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>VFMI, dysphagia</td>
<td>1</td>
</tr>
<tr>
<td>Pourmoghadam29</td>
<td>Yes</td>
<td>2</td>
</tr>
<tr>
<td>Pham12</td>
<td>VFMI, dysphagia</td>
<td>Yes</td>
</tr>
<tr>
<td>Skinner13</td>
<td>Yes</td>
<td>3</td>
</tr>
<tr>
<td>Mery27</td>
<td>VFMI</td>
<td>Yes</td>
</tr>
<tr>
<td>Kohr30</td>
<td>VFMI, dysphagia</td>
<td>Yes</td>
</tr>
<tr>
<td>LoTempio31</td>
<td>Yes</td>
<td>4</td>
</tr>
<tr>
<td>Averin28</td>
<td>VFMI, dysphagia</td>
<td>Yes</td>
</tr>
<tr>
<td>Alfares17</td>
<td>Yes</td>
<td>5</td>
</tr>
</tbody>
</table>

Abbreviation: VFMI, vocal fold motion impairment.


All studies were retrospective chart reviews and conducted in the United States.

Records identified through database searching (n = 1880) → Additional records identified through other sources (n = 2) → Total records identified (n = 1882) → Duplicates removed (n = 872) → Records screened (n = 1008) → Records excluded (n = 956) → Full-text articles assessed for eligibility (n = 52) → Full-text articles excluded, with reasons (n = 44) → Studies included in qualitative synthesis (n = 8)

Figure 1. Flow diagram of the literature search based on the PRISMA recommendations (Preferred Reporting Items for Systematic Reviews and Meta-analyses). Patient Selection and Characteristics
Demographic characteristics for the studies meeting inclusion criteria are outlined in Table 1. The most commonly performed procedures were Norwood procedures and aortic arch reconstruction, but the studies in this review also included subjects who underwent a Blalock-Taussig shunt, Fontan procedure, Sano shunt, atrial septal defect repair, ventricular septal defect repair, repair of valves, ligation of PDA, and others. Studies that included patients who underwent isolated PDA ligation were excluded. The average age of subjects in most studies was <1 month, but in 2 studies the average was >1 year. One study did not report the average age of its subjects. Of the 5 studies that reported weights for their subjects, 4 had similar means and distributions of weight. With regard to patients with genetic syndromes, 3 studies included, 2 studies excluded, and 3 studies made no comment regarding exclusion or inclusion of these subjects. Follow-up was highly variable, ranging from 6 months to 6 years. Mortality rates were reported in 3 studies, and none suggested an association to VFMI; however, only 1 study reported mortality rates stratified by the presence or absence of VMFI. Data were analyzed according to the availability and specificity of data related to VFMI and dysphagia outcomes.

**Bias Assessment**

The US National Institutes of Health’s Quality Assessment of Case Series Studies tool was used to assess the risk of bias in the included studies (Table 2). With the exception of the study by LoTempio and Shapiro,31 the included studies appeared to have similar objectives and were executed in comparable fashion. The study by LoTempio and Shapiro was a case series, and the data were primarily qualitative, which did differ from the other studies, most of which were retrospective cohorts and reported quantitative data. The 2 categories that had the highest risk of bias in this study were item 6 (Were the outcome measures clearly defined, valid, reliable, and implemented consistently across all study participants?) and item 7 (Was the length of follow-up adequate?). In the 4 studies that did not satisfy item 6, there was a considerable amount of variability in the outcome measures. One of the most apparent examples of this was that some of the studies employed a symptom-driven screening protocol, whereas others used a universal screening protocol for both VFMI and dysphagia. This is discussed later in this review. The mean length of follow-up was <1 year in 3 studies, and 3 studies did not report the length of follow-up times. The remaining items addressed in the bias assessment were satisfied in the majority of the studies.

**VFMI Outcomes**

The incidence of VFMI ranged from 8% to 59% of subjects in the 8 studies reporting outcomes on subjects with VFMI after cardiac surgery (Table 3). The study with the symptom-driven protocol (Kohr et al30) had the lowest incidence of VFMI (8%) as compared with studies with a universal VFMI screening protocol (10%-59%). All except 1 study reported that unilateral VFMI, particularly left-sided VFMI, made up the majority of subjects with VFMI. Averin et al38 noted that their subject with right-sided VFMI had undergone a right-sided aortic arch reconstruction. The remaining studies with right-sided unilateral VFMI did not remark on any unusual circumstances that may have explained this finding. Six studies employed a universal VFMI screening where all subjects had flexible nasolaryngoscopy (FNL) performed postoperatively; 1 study had a symptom-driven protocol for postoperative FNL evaluation; and 1 study did not specify what protocol it used.

Data on recovery of VFMI based on follow-up FNL evaluations were reported in 5 studies. Resolution of VFMI ranged from 8% to 96% at 6 months to 6 years of follow-up. Partial improvement was reported in 2 studies and ranged from 25% to 26% at 6 to 12 months. Of the studies with <1 year of mean follow-up time, resolution of VFMI ranged from 8% to 60%, whereas the studies that had >1 year of mean follow-up time reported rates of resolution between 61% and 96%. Alfares et al17 found that the median time to recovery was 10 months (range, 1-48 months) among subjects who recovered function, with a median follow-up of 385 days (range, 1 month–3 years). All studies reporting rates of recovery or improvement in VFMI employed a universal screening protocol.

**Respiratory Outcomes**

Ventilatory support data were reported in 4 of the included studies, and 3 of these studies compared data from subjects with VFMI with those without or the total cohort. Subjects with VFMI in the studies by Kohr et al30 and Averin et al17 spent more time on a ventilator on average as compared with subjects without VFMI, while the opposite was found in the study by Pourmohaghadam et al.29 However, there was no significant difference in the time spent on the ventilator overall in the 2 populations. In addition, there was some variability in hospital length of stay in subjects with VFMI among studies. Once again, however, these differences did not show any meaningful significance. Tracheotomy outcome data were sparse in this data set. Alfares et al17 and Pham et al12 reported incidence of tracheotomy placement at 3 of 31 and 10 of 151 in their respective cohorts. Alfares et al also reported that 7 of 31 subjects underwent “additional airway procedures,” though it was not specified what these procedures were. The Alfares et al study population consisted only of subjects with VFMI without a comparison group, and Pham et al did not specify who among the subjects who had tracheostomies also had VFMI. Therefore, it is not clear how much VFMI status influences the need for a tracheotomy or additional airway procedures. Bilateral VFMI was present in 2 of 3 subjects requiring a tracheotomy in the Alfares et al study, and this is consistent with other studies that reported a higher rate of tracheotomy with after CHS.16,19 LoTempio and Shapiro31 reported initial successful decannulation in 2 of 6 subjects with VFMI and tracheostomies; however, 1 required replacement of the tracheotomy during the long-term follow-up period. Voice
### Table 2. Demographic Characteristics of Included Studies.

<table>
<thead>
<tr>
<th>First Author</th>
<th>Publication Year</th>
<th>Age</th>
<th>Weight</th>
<th>Syndromic Patients</th>
<th>Type of Surgery</th>
<th>Overall Mortality</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pourmoghadam</td>
<td>2017</td>
<td>13 (15) d</td>
<td>3.2 (0.6) kg</td>
<td>Not reported</td>
<td>Norwood, 63; non-Norwood arch reconstruction, 26</td>
<td>VFMI +, 2 of 44; VFMI −, 4 of 46</td>
<td>11 mo; 3.2 y among patients with dysphagia</td>
</tr>
<tr>
<td>Pham</td>
<td>2014</td>
<td>12 (7-21) d</td>
<td>Not reported</td>
<td>Yes</td>
<td>Norwood, 65; AA reconstruction, 39</td>
<td>55 of 151</td>
<td>12 mo</td>
</tr>
<tr>
<td>Skinner</td>
<td>2005</td>
<td>Norwood, 6 (2-19) d; AA reconstruction, 9 (4-33) d</td>
<td>Norwood, 3.1 (1.9-4.2) kg; AA reconstruction, 3.1 (2.0-3.7) kg</td>
<td>Not reported</td>
<td>Norwood, 33; AA reconstruction, 18</td>
<td>Not reported</td>
<td>6 mo</td>
</tr>
<tr>
<td>Mery</td>
<td>2014</td>
<td>Not reported</td>
<td>Not reported</td>
<td>Yes</td>
<td>AA advancement with additional procedures, 275</td>
<td>Not reported</td>
<td>6 y</td>
</tr>
<tr>
<td>Kohr</td>
<td>2003</td>
<td>4.8 (5.2) y (1 d–17 y)</td>
<td>20 (18, 3.1-73) kg</td>
<td>No</td>
<td>ToF repair; VSD repair; stage I Norwood; Fontan procedure; AA reconstruction; arterial switch</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
<tr>
<td>LoTempio</td>
<td>2002</td>
<td>30 (25) mo</td>
<td>Not reported</td>
<td>Not reported</td>
<td>Coarctation repair; 2; coarctation repair with PDA, 1; BTS, 1; valve repair, 2</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
<tr>
<td>Averin</td>
<td>2012</td>
<td>6 d</td>
<td>3.1 (0.6) kg</td>
<td>No</td>
<td>Norwood, 63</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
<tr>
<td>Alfares</td>
<td>2015</td>
<td>VFMI recovery, 21 (7-41) d; persistent VFMI, 11 (6-24) d</td>
<td>VFMI recovery, 2.6 (1.2-3.6); persistent VFMI, 2.7 (1.1-3.6)</td>
<td>Yes</td>
<td>PDA ligation, 22; AA reconstruction, 29; Norwood stage I, 5; VSD repair, 4; ASD repair, 4; BTS, 2; Sano shunt, 2; repair of TAPVR, 1</td>
<td>2 of 29</td>
<td>Not reported</td>
</tr>
</tbody>
</table>

**Abbreviations:** AA, aortic arch; ASD, atrial septal defect; BTS, Blalock-Taussig shunt; PDA, patent ductus arteriosus; TAPVR, total anomalous pulmonary venous return; ToF, tetralogy of Fallot; VFMI, vocal fold motion impairment; VSD, ventricular septal defect.

*Mean (SD).*

*Mean.*

*Mean (SD, range).*

*Median (range).*

*Median.*

*Median (interquartile range).*
assessment data were scarce; however, Skinner et al\textsuperscript{13} reported that a weak cry was noted in 5 of 7 subjects with VFMI on initial assessment, and this was statistically significant. Meaningful data on long-term voice outcomes were not reported.

### Dysphagia Outcomes

The incidence of dysphagia and need for tube feeding ranged from 16\% to 76\% across the 5 studies reporting swallowing outcomes (Table 4). Higher incidences of clinically apparent dysphagia (on modified barium swallow [MBS] or fiberoptic endoscopic evaluation of swallowing) or enteral feeding supplementation (nasogastric [NGT] or gastric tube [GT] feeding) in subjects with VFMI as compared with those without VFMI were demonstrated in 4 of 5 studies, whereas the remaining study did not find any difference between the groups. A universal dysphagia screening where all subjects underwent a formal feeding evaluation with an MBS or fiberoptic endoscopic evaluation of swallowing was employed in 3 of 5 studies, whereas studies with universal screening protocols reported incidences between 16\% and 62\%. Of the 2 studies that did not have universal dysphagia screening protocols, 1 found a higher incidence of dysphagia in subjects with VFMI, whereas the other did not find any difference between the groups.

The inability to maintain an oral diet was present in 14\% to 100\% of subjects with VFMI and 11\% to 61\% of subjects without VFMI following surgery. The majority of subjects, with and without VFMI, were eventually able to recover their swallowing function. Overall, 66\% to 75\% of subjects with VFMI and 88\% to 100\% of subjects without VFMI were able to tolerate an oral diet without tube feeding at 23.6 days to 3.2 years of follow-up. Given the wide range of follow-up time among studies, it was difficult to accurately determine the time to resolution. Kohr et al\textsuperscript{30} evaluated the length of time of NGT use for subjects with dysphagia, and their results indicate that most cases of swallowing dysfunction resolved within several months of surgery. Similarly, Skinner et al\textsuperscript{13} found evidence of aspiration on MBS in 14 subjects; on repeat MBS at follow-up, only 2 demonstrated trace aspiration, and 3 demonstrated laryngeal penetration. Follow-up for this study was 6 months, and so this also provides evidence to suggest that dysphagia resolves relatively quickly.

In many of the studies, it is unclear whether some of these subjects were able to tolerate an oral diet but still had some dependence on tube feedings. Pourmoghadam et al\textsuperscript{29} reported that among the subjects with VFMI, 8 of 42 were able to tolerate an oral diet but still required tube feeds, and 34 of 42 were entirely tube feed dependent at the time of discharge. At follow-up, only 4 of 41 were tube feed dependent. This is in contrast to the subjects without VFMI, where 12 of 45 were tube feed dependent at discharge, but no subjects were requiring tube feeds at the last follow-up.

Additional interventions related to swallowing were not common except in the study by Pourmoghadam et al.\textsuperscript{29} In that study, Nissen procedures were performed more frequently in subjects with VFMI. Skinner et al\textsuperscript{13} also reported 2 subjects having a Nissen procedure, but neither subject had VFMI.

### Discussion

Our systematic review evaluated the relationship between VFMI and both respiratory complications and dysphagia.
<table>
<thead>
<tr>
<th>First Author</th>
<th>Subjects, n</th>
<th>UVFMI / BVFMI</th>
<th>Ventilatory Support, d</th>
<th>LOS Outcomes, d</th>
<th>Rate of Recovery Outcomes</th>
<th>NPL Protocol</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mery(^{27})</td>
<td>VFMI +, 36; VFMI −, 59</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>VFMI recovery: 35 of 36 based on clinical symptoms only (no follow-up NPL)</td>
<td>Universal</td>
</tr>
<tr>
<td>Kohr(^{10})</td>
<td>VFMI +, 4; VFMI −, 46</td>
<td>UVFMI, 4</td>
<td>VMFI +, 8 (7, 1-17); all patients, 2.5 (3.4)(^{b})</td>
<td>VMFI +, 12 (7.5-22); VMFI −, 17 (4.9, 13-25)(^{a})</td>
<td>NR</td>
<td>Symptom driven</td>
</tr>
<tr>
<td>LoTempio(^{11})</td>
<td>VFMI +, 6; VFMI −, 90</td>
<td>UVFMI, 2; R VFMI, 1; BVFMI, 3</td>
<td>15 (20, 0-50)(^{a})</td>
<td>NR</td>
<td>Tracheostomy: 2 of 6 decannulated; 4 of 6 tracheostomy dependent</td>
<td>NR</td>
</tr>
<tr>
<td>Averin(^{28})</td>
<td>Normal VF motion, 26; VFMI, 37</td>
<td>UVFMI, 35; R VFMI, 1; BVFMI, 1</td>
<td>VFMI +, 8 (4.8-11); VFMI −, 5.8 (4.6-9.9)(^{d})</td>
<td>VFMI +, 23.5 (18-33.5); VFMI −, 24 (17-38)(^{d})</td>
<td>NR</td>
<td>Universal</td>
</tr>
<tr>
<td>Pourmoghadas(^{29})</td>
<td>VFMI +, 43; VFMI −, 58</td>
<td>UVFMI, 41; R VFMI, 1; BVFMI, 1</td>
<td>VFMI +, 9 (0.3); VFMI −, 12 (0.4)(^{b})</td>
<td>VFMI +, 45 (35-64); VFMI −, 39 (29-64)(^{d})</td>
<td>VFMI recovery: 14 of 26 in Norwood group; 12 of 17 in non-Norwood group</td>
<td>Universal</td>
</tr>
<tr>
<td>Pham(^{12})</td>
<td>VFMI +, 60; VFMI −, 44</td>
<td>UVFMI, 56; R VFMI, 2; BVFMI, 2</td>
<td>NR</td>
<td>NR</td>
<td>VFMI recovery: 15 of 23 with no improvement; 6 of 23 with partial improvement; 2 of 23 with resolution</td>
<td>Universal</td>
</tr>
<tr>
<td>Skinner(^{13})</td>
<td>VFMI +, 7; VFMI −, 47</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>VFMI recovery: 1 of 7 with improvement; 3 of 7 without follow-up NPL</td>
<td>Universal</td>
</tr>
<tr>
<td>Alfares(^{17})</td>
<td>VFMI, 31</td>
<td>BVFMI, 5; R VFMI, 4; LVFMI, 22</td>
<td>NR</td>
<td>VF recovery, 51 (33-108); persistent VFMI, 45 (31-98)(^{d})</td>
<td>VFMI recovery: 19 of 31 recovered with median recovery time of 10 mo</td>
<td>Universal</td>
</tr>
</tbody>
</table>

Abbreviations: BVFMI, bilateral vocal fold motion impairment; ICU, intensive care unit; LOS, length of stay; NPL, nasopharyngolaryngoscopy; NR, not reported; UVFMI, unilateral vocal fold mobility impairment; VF, vocal cord; VFMI, vocal fold motion impairment.

\(^{a}\)Mean (SD, range).

\(^{b}\)Mean (SD).

\(^{c}\)Subject had right-sided aortic arch reconstruction.

\(^{d}\)Median (interquartile range).
following CHS. We also examined long-term recovery of VFMI and swallowing dysfunction. Overall, the existing literature demonstrated significant heterogeneity. Both symptom-driven and universal screening protocols were utilized, with incidence of VFMI ranging from 8% to 59%, while recovery of motion ranged from 8% to 96%. Respiratory outcomes were extremely variable, with a small proportion of patients requiring tracheotomy. The relationship between need for tracheotomy and VFMI was poorly described through studies. Dysphagia incidence ranged from 16% to 76% across the included studies, and it appeared that there was a correlation between development of dysphagia and VFMI. However, recovery of swallowing function and vocal fold motion appeared to be independent of each other across multiple studies.

As the mortality rate associated with CHS has improved over time, there has been a greater focus on managing the postoperative complications and morbidity associated with CHS. Although not common, injury to the RLN is a well-established risk of CHS and is regarded as a significant risk factor for postoperative morbidity and mortality. Because the RLN innervates most of the intrinsic muscles of the larynx and carries some of the sensory afferent fibers from the cervical esophagus and trachea, injury to the RLN could result in a compromised airway and dysphagia. In the postoperative period following CHS, nutrition and airway protection are particularly important given the already fragile status of the neonate patient. Early diagnosis and intervention with speech and swallow therapy may be essential for the prevention of future complications and may help to avoid invasive procedures for airway protection and maintenance of nutrition. Many patients with VFMI are asymptomatic and may continue to be asymptomatic in the long term despite still having VFMI; however, a considerable proportion are very symptomatic and will continue to be affected over the long term.

Significant rates of swallowing dysfunction were seen in patients with and without VFMI. Most studies demonstrated a higher rate of dysphagia in subjects with VFMI as compared with subjects without VFMI in this review as well as other similar studies and so a finding of VFMI on FNL should raise suspicion for swallowing dysfunction. However, given the significant proportion of patients with dysphagia without VFMI, clinicians should maintain a high index of suspicion and consider swallow evaluations in all patients. Recovery of swallowing dysfunction and VFMI were highly variable in this study. Overall, the majority of subjects with swallowing dysfunction postoperatively were able to tolerate an oral diet during the course of their follow-up. The rates of successful diet advancement were higher in subjects without any apparent VFMI but still very favorable even among the subjects with VFMI.

The data on recovery of VFMI was less consistent as compared with the data on swallowing dysfunction. The rate of recovery ranged from 8% to 96%, but most of the studies reported that many of the subjects had at least improvement or partial resolution. The follow-up periods for these studies were fairly short overall, and this may explain why the rates of recovery appear very poor in some studies. The follow-up period in the study by Mery et al was an average of 6 years, and the recovery rate in this study was 96%. The major flaw of this study is that recovery was based on clinical improvement, and there was no routine follow-up flexible laryngoscopy to assess recovery of motion. It is clear, based on these studies, that swallowing recovery occurs independent of VFMI; therefore, the concept of recovery based on clinical improvement should be viewed with caution. This is in contrast to the other studies that examined recovery of VFMI, where repeat flexible laryngoscopy was performed and the rates of recovery were much lower. It is unclear how symptomatic the subjects with VFMI were long term or if they were able to adequately compensate. Data on the need for tracheostomies in these patients were extremely sparse, and there was only 1 study included in this review that reported rates of tracheostomies in this patient population, although the study did not specify how many subjects who needed a tracheotomy had VFMI. Based on the limited data, a minority of subjects required a tracheotomy following CHS. However, among subjects who had received a tracheotomy, most did not undergo decannulation within the follow-up period. There were no data reported on rates of laryngoplasty, reinnervation, or other airway procedures aside from tracheotomy. VFMI status did not appear to have an effect on mortality. These data highlight the fact that the presence of VFMI is only one of the key players in morbidity and mortality after CHS. The general health of the patient, the type of congenital heart disease, and other underlying airway issues also play a role in overall outcomes. Further prospective work should account for these confounders when determining the overall impact of VFMI on the patient’s overall status.

The importance of assessing for VFMI postoperatively is supported by our finding that there are higher rates of a VFMI diagnosis in studies that have a universal FNL screening protocol as compared with those that are symptom driven. This is consistent with findings from a large meta-analysis that in part examined rates of VFMI diagnosis after PDA ligation and CHS. Most studies do not routinely assess for VFMI preoperatively, and so it is unclear what the actual risk is to the RLN due to CHS. One study did perform FNL evaluations on neonatal patients prior to CHS or PDA ligation and found that the incidence of VFMI preoperatively was 2.5% and postoperatively was 8%, supporting the assumption that most cases of VFMI following CHS are iatrogenic in etiology.

The current data available are limited to retrospective studies with fairly short-term follow-up periods. Data heterogeneity complicated our identification of eligible studies and data analysis and precluded the option of performing a meta-analysis. We chose to exclude PDA ligation data from our study because the literature indicates that this population is potentially different from the CHS population, including a large premature population that may not have any other
true congenital heart disease. In a meta-analysis, Strychowski et al found that subjects who had undergone PDA ligation had higher rates of VFMI as compared with the entire cohort of subjects who had undergone any type of cardiac surgery. Many publications examining the outcomes of VFMI and swallowing dysfunction following CHS pool data with a PDA ligation cohort without reporting results stratified by the type of surgery. Unfortunately, this resulted in many studies identified on our initial query being excluded from this review; however, excluding studies that included PDA ligation resulted in a more comparable study population than prior studies. In addition, it is possible that the included studies included patients who had VFMI as a result of cricoarytenoid joint fixation secondary to prolonged intubation, especially in this particular population. Of the studies that were included, the populations differed in types of surgery, ages, surgical weights, and whether nonsyndromic subjects were included. Another notable limitation lies in the fact that a number the included studies do not stratify outcomes by unilateral VFMI and bilateral VFMI, which obviously manifest differently. Although the majority of cases of VFMI were unilateral, it is certainly likely that including bilateral VFMI does after some findings, related to both swallowing and respiratory outcomes. Last, we have to acknowledge that different surgeons may have different operative methods and skill levels that may affect postoperative outcomes. This systematic review highlights the need for prospective studies that evaluate the effect of universal vocal fold motion, voice, and swallow evaluations pre- and postoperatively. This will help us identify patients at highest risk of VFMI and dysphagia after CHS, allowing for generation of a targeted screening algorithm, appropriate resource utilization postoperatively, and improved short- and long-term outcomes.

Conclusion

Dysphagia and VFMI are known risks of CHS and have the potential to cause significant morbidity in the postoperative period. This systematic review demonstrated that the majority of subjects with dysphagia following CHS will recover significant function; however, recovery of VFMI was less predictable. Further studies, particularly prospective studies, would greatly enhance our ability to identify high-risk populations, intervene when appropriate, and predict the probability of recovery.

Author Contributions

Susannah Orzell, acquisition, interpretation, analysis, revising and accountability; Rahul Joseph, acquisition, analysis, revising and accountability; Julina Ongkasuwan, interpretation, analysis, revising and accountability; Joshua Bedwell, interpretation, analysis, revising and accountability; Jennifer Shin, interpretation, analysis, revising and accountability; Nikhila Raol, design, acquisition, analysis, drafting, and accountability.

Disclosures

Competing interests: Julina Ongkasuwan, Springer Publishing—royalties; Jennifer Shin, Springer Publishing—book royalties for Evidence-Based Otolaryngology; Plural Publishing—book royalties for Otolaryngology Prep and Practice; Harvard Medical School—Shore Foundation Faculty Award; Brigham and Women’s Hospital—Care Redesign Award. 

Funding source: None.

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