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Retrospective Study

Outcomes of Vocal Fold Immobility After Pediatric Cardiovascular Surgery

Anatoli F. Karas, MD[1]; Aniruddha Patki, MD[1]; Marisa A. Ryan, MD, MPH[1]; Patrick A. Upchurch, MD[2]; Rose J. Eapen, MD[1]; Eileen M. Raynor, MD[1*]

[1]Department of Surgery, Division of Head and Neck Surgery & Communication Sciences, Duke University Medical Center, DUMC 3805, Durham, NC 27710, USA
[2]Department of Surgery, Boston Children's Hospital, 300 Longwood Avenue, Fegan 3, Boston, MA 02115, USA

[*]***Corresponding author***
Eileen M. Raynor, MD
Associate Professor, Department of Surgery, Division of Head and Neck Surgery & Communication Sciences, Duke University Medical Center, DUMC 3805, Durham, NC 27710, USA; Tel. 919-613-4573; Fax. 919-668-3103; E-mail: eileen.raynor@duke.edu

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ABSTRACT

Objectives
Cardiovascular surgery is increasingly performed in children for congenital malformations of the heart and great vessels. Observed as vocal fold immobility, recurrent laryngeal nerve injury is a well-described complication. As overall outcomes improve and patients live longer, the sequelae of vocal fold immobility amplify insignificance. Families are often unaware of the long-term issues related to vocal fold immobility especially with regard to need for alternative alimentation routes. We report on the incidence of use of feeding tubes, the timing of vocal fold function return and need for additional aerodigestive interventions.

Methods
We reviewed 65 patients <2-years who underwent cardiovascular surgery at a tertiary center from 2008-2013 and were diagnosed post-operatively with vocal fold immobility by fiberoptic examination.

Results
Patent ductus arteriosus and hypoplastic left heart syndrome were the most common of the cardiovascular anomalies included. The majority (92%) had unilateral left immobility. Recovery of motion was observed in 29% ranging from 1-month to 3-years. Hypoplastic left heart syndrome had a significant negative correlation with recovery. Forty-six percent required gastrostomy tube placement either to supplement their oral intake or to completely meet their nutritional needs. Forty-eight percent required subsequent aerodigestive surgery including direct laryngoscopy/bronchoscopy (22%), tracheostomy (8%) and vocal fold injection (5%).

Conclusion
Counseling of families and primary care providers regarding the impact of pediatric vocal fold immobility after cardiac surgery should include the high potential requirement for supplemental alimentation as well as the need for feeding and speech therapy. Longitudinal otolaryngology and speech pathology care is imperative with vocal fold immobility since the majority of these patients do not experience functional recovery and may require further interventions over time.

Keywords
Vocal cord paralysis; Pediatric cardiovascular surgery; Pediatric aerodigestive disorders; Voice; Swallow; Gastrostomy.

INTRODUCTION

Cardiovascular surgery is increasingly performed in infants to correct congenital malformations of the heart and great vessels. Due to the anatomic course of the recurrent laryngeal nerve, it is especially prone to injury during procedures requiring aortic arch manipulation and/or reconstruction. These malformations range in both incidence and complexity. Patent ductus arteriosus (PDA) represents up to 10% of all congenital heart disease with an incidence of up to 2.8 per 1000 live births.[1] Surgical PDA ligation

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can be performed without cardiopulmonary bypass and requires limited dissection around the aortic arch. Conversely, hypoplastic left heart syndrome (HLHS) is a single ventricle lesion that occurs in approximately 0.18 per 1000 live births and requires staged palliation and extensive aortic arch reconstruction under deep hypothermic circulatory arrest or regional cerebral perfusion.[2]

Observed post-operatively as vocal fold immobility, recurrent laryngeal nerve injury is a well-described complication of these surgeries. Prolonged or traumatic intubations, which can also occur in this patient population, can also cause post-extubation vocal fold immobility. The reported incidence of vocal fold immobility after pediatric cardiovascular surgery varies in the literature from 8.7% to 39%.[3] The use of extracorporeal membranous oxygenation (ECMO) also increases the risk of recurrent laryngeal nerve (RLN) injury, primarily on the right side.[4] Multiple factors have been reported to predispose to vocal fold immobility, including low birth weight, younger age at surgery, and use of electrocautery during dissection around the aorta.[5-7] However, data is lacking on long-term outcomes of vocal fold immobility in pediatric patients after cardiovascular surgery. As techniques and overall outcomes improve, these patients live longer and the sequelae of vocal fold immobility amplify in significance. We report on the incident need for supplemental alimentation *via* surgical feeding tube, the timing of vocal fold motion return and the requirement for additional aerodigestive interventions in a series of these children from a single tertiary academic medical center.

METHODS

Population

We performed a retrospective chart review of all 65 patients <2-years-old who underwent cardiovascular surgery at Duke University Medical Center (DUMC) from 2008 to 2013 and were diagnosed post-operatively with vocal fold immobility by awake, bedside fiberoptic examination (FOE). The FOE was performed by otolaryngology resident and viewed by an otolaryngology attending either directly or on video recording.

Inclusion criteria were: less than 2-years-old at the time of their first corrective cardiovascular surgery; primary surgery performed at Duke and assigned international classification of diseases-9 (ICD-9) codes for vocal fold immobility and congenital cardiovascular abnormality. At DUMC, a diagnosis of vocal fold immobility is only made by otolaryngology and, therefore, all included patients were evaluated by the otolaryngology service.

Data Collection and Analysis

Patient charts were reviewed to determine the date of surgery and dates of subsequent follow-up with otolaryngology. Follow-up was defined as any inpatient evaluation or outpatient clinic visit, regardless of whether or not FOE was performed. The dates of post-operative follow-up visits were measured from the patient's surgery and used to determine the time to recovery. Recovery was defined by either a FOE documenting the return of vocal fold mobility, or documentation of a normal cry, completely oral feeding status, and breathing if FOE was not performed.

Secondary measures collected included the dates and types of operative procedures on the airway, including direct laryngoscopy/bronchoscopy, vocal fold injection, and tracheostomy. We also determined the proportion of patients requiring surgical placement of a feeding tube. The numbers in each category were evaluated with the Pearson's Chi-squared test for count data (more than two categories) assuming equal probability for each category. The significance level for assessing the statistical test was set to α=0.05. All analyses were conducted using excel 2010. This study was approved by the Duke Institutional Review Board (Pro00045147).

RESULTS

A total of 65 patients met the inclusion criteria. Of these, 18 patients had a patent ductus arteriosus (PDA), 17 had HLHS, 13 had aortic coarctation, 12 had an interrupted aortic arch, two had transposition of great vessels, one had pulmonary atresia, one had atrioventricular canal defect and one had Tetralogy of Fallot. The median follow-up was 131-days after surgery. The majority of patients (92%) had unilateral immobility of the left vocal fold. There were also four cases of right vocal fold immobility and one with bilateral vocal fold immobility.

All diagnoses of vocal fold immobility were made by FOE while still an inpatient after their initial cardiovascular surgery in all cases. The most common indication for otolaryngology consult was a weak cry following extubation. At DUMC, if there is a clinical concern for vocal fold immobility, the Pediatric Cardiothoracic Intensive Care Unit will withhold oral feeding until after the vocal fold function is assessed, so swallow function was very infrequently known prior to the initial FOE.

Aerodigestive Interventions

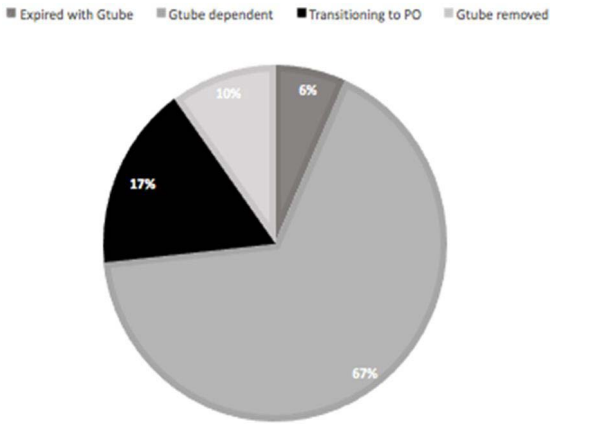
During subsequent follow-up, the majority of patients (58%) only underwent one additional FOE. However, one underwent none, 14 (21%) had two, eight (12%) had three, three (4%) had four, and a single patient had five. The most common indication for FOE was it being the first outpatient examination since the diagnosis of vocal fold immobility was made while an inpatient. Another common indication was plateauing of symptoms, such as continued difficulty with oral intake or continued weak cry. The third reason for performing FOE was parental request or anxiety about the prior diagnosis of immobility. The most common reason for not performing a FOE in the office was that parents reported significant improvement in swallowing ability or cry. A less common reason for deferring FOE in the office was an upcoming surgical procedure for the child with a plan to perform direct laryngoscopy/bronchoscopy in the operating room.

Out of all the patients, 31 (48%) underwent a secondary surgical procedure either involving the airway or for a surgical feeding tube placement. The most common airway procedure

performed was direct laryngoscopy/bronchoscopy performed in 14 (22%) patients. Eight of those 14 underwent a single direct laryngoscopy/bronchoscopy whereas four underwent two direct laryngoscopy/bronchoscopies and two patients underwent three direct laryngoscopy/bronchoscopies. Placement of a tracheostomy was necessary for 5 (8%) patients. It was performed for airway obstruction in the setting of bilateral vocal fold paralysis in one patient, respiratory failure in the setting of laryngomalacia and left vocal fold immobility in one patient, tracheobronchomalacia in one patient and ventilator dependence in two patients. Injection medialization of the immobile vocal fold was performed in three (5%) patients.

Surgical placement of a feeding tube was necessary for 30 (46%) of patients. Three of these patients had a surgical feeding tube placed prior to otolaryngology consultation. The most common indication for feeding tube placement was severe reflux in 17 (58%) followed by feeding intolerance in 12 (41%). One patient required a surgical feeding tube for failure to thrive. All of the patients were seen by speech pathology for feeding evaluation and treatment. Of the patient's with surgical feeding tubes, 14 (46%) had at least one videofluoroscopic swallow study (VFSS) for feeding evaluation during initial admission or in follow-up as an outpatient. Seven (23%) of the patients had VFSS prior to surgical feeding tube placement. Four of these showed aspiration. The other three showed no aspiration but continued with gastrostomy tube placement secondary to oral feeding intolerance. Of patients with surgical feeding tubes, 5 (17%) were noted to be transitioning to full oral feeds but their feeding tube remained in place at the end of the study period. Three (10%) children had their surgical feeding tube ultimately removed at a mean of 16-months after placement. Twenty (67%) remained dependent on the feeding tube with little or no significant oral intake at the end of the study period and 2 (7%) expired with the feeding tube in place (Figure 1). Of the 31 patients who underwent subsequent airway procedures or feeding tube placement, 19% exhibited vocal fold recovery. In comparison 13/34 (38%) of children who did not require a second surgical procedure to address their airway, voice or feeding had vocal fold movement recovery. This was not a statistically significant different distribution (p=0.09).

Figure 1. *Feeding Tube Status by End of Study Period*



Recovery of Vocal Fold Mobility

Among the 65 patients, 19 (29%) exhibited recovery of vocal fold mobility while 46 (71%) did not. Of the 19 patients with documented return of vocal fold mobility, eight (42%) were based on FOE and 11 (58%) were based clinically on return of normal cry and completely oral feeding status. Among the 19 patients who had recovered vocal fold motion, time to recovery varied widely, with a median of 6-months and range between 1-month to 3-years (Figure 2). In the patients with return of vocal fold motion, 14 (74%) had return of motion documented by 1-year. When patients were stratified by diagnosis, only HLHS exhibited a statistically significant negative correlation with recovery; only one of the 17 patients with HLHS exhibited recovery whereas the remaining 16 did not (Figure 3). This distribution was significantly different from the remainder of the patients (p=0.03).

Figure 2. *Days After Cardiovascular Surgery to Recovery of Vocal Fold Movement with Quartiles*

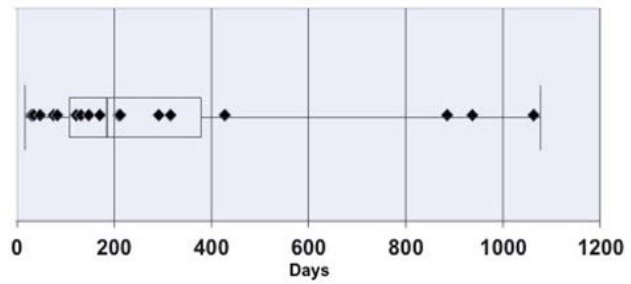
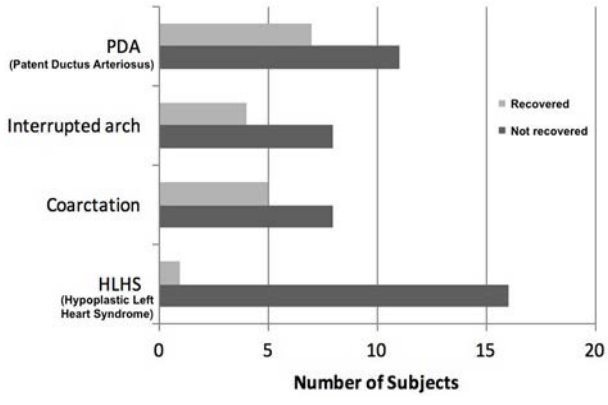


Figure 3. *Recovery of Vocal Fold Movement Stratified by Cardiovascular Diagnosis*



DISCUSSION

Iatrogenic injury to the recurrent laryngeal nerve is the most likely presumed etiology of vocal fold immobility after cardiovascular surgery, but other factors that may contribute include extended periods of intubation, vagal injury from ECMO cannulation or underlying neurologic abnormality. Wilson et al reported that younger age, complex structural abnormality as opposed to a sim-

pler PDA, need for pre-operative intubation, median sternotomy with delayed closure, and palliative surgery were all associated with longer intubation times following pediatric cardiothoracic surgery.[8] In our series, 47/65 (72%) of patients underwent cardiovascular surgery for a structural cardiac defect other than PDA. Furthermore, 17 of these patients underwent aortic arch reconstruction as part of the staged palliation of HLHS. A high proportion of our patients were therefore at risk for prolonged intubation as they recovered from their cardiovascular surgery. Since FOE for vocal fold mobility can only be performed after the child is extubated, it is possible that intubation-related injury was a contributing factor to cases of vocal fold immobility in our study. It was not possible from a retrospective review to differentiate between these potential causes. However, the significant preponderance of left-sided vocal fold paralysis in this study suggests that intra-operative recurrent laryngeal nerve injury was the most common etiology, as we would expect intubation injury, ECMO or an underlying neurologic problems to affect both vocal folds more equally.

Recovery of Function

In this series, 19 patients (29%) exhibited recovery of function over the duration of follow-up. This is a similar to previously reported recovery of 28% and 35%.[9,10] We found a median time to recovery of 6-months with a wide range from 1-month to 3-years (Figure 1). This median is slightly longer than the 130-days found by Jabbour et al.[10] Of the 19 patients who had recovery of function, 13 achieved this by 7-months after their cardiovascular surgery. The remaining six patients exhibited recovery over 1-year from their cardiovascular surgery. Although recovery this long after surgery has been reported, these patients may have actually regained vocal fold movement earlier, but it was documented at a later date when the next clinical follow-up occurred. This measurement error likely falsely increased the recorded duration to recovery for some of the subjects as well as the median time to recovery. An additional limitation of the retrospective nature of this study is that 11 (58%) of the vocal fold mobility recoveries were based clinically on voice and swallow normalization and not visually confirmed with FOE. Some of these symptom-based recoveries may be due to a medially positioned immobile vocal fold or compensatory functions. Therefore, our overall incidence of recovery may be artificially high without confirmatory FOEs on all patients. It should be noted that parents often decline follow-up FOE if the patient is asymptomatic or has improved, which is one reason not all recoveries were documented with FOE.

Recovery in Patients with Hypoplastic Left Heart Syndrome

In our analysis, the only predictive factor for recovery of vocal fold movement was the negative association of recovery with a diagnosis of HLHS. This may be due to several reasons. First, patients with HLHS undergo multiple staged procedures as part of their treatment. These are generally performed around 1-week of life, 4-6-months-old, and 3-4-years-old. There is a very wide range of incidence of vocal fold immobility reported after the first stage, from 9% to 58.7%.[6,11] The method of aortic dissection used may explain some of this variation as Averin et al reported a significant drop in the incidence of vocal fold immobility after this procedure when their group started using a blunt dissection technique rather than electrocautery.[6] Incidence of vocal fold paralysis after the 2nd and 3rd staged procedures has not been well reported, but it is plausible that repeat surgical procedures in the chest may increase the risk of vocal fold immobility. Additionally, although the average period of mechanical ventilation is between three and seven days following the first stage of Norwood reconstruction,[12] potential acute post-operative complications including drastic lability in pulmonary blood flow, derangement of coronary blood flow, myocardial edema, myocardial dysfunction, seizures, stroke, chylothorax, and phrenic nerve injury could contribute to prolonged periods of intubation. It is also possible that the severity of congenital heart disease in these children relegates the sequelae of vocal cord immobility to a lower priority. Consequently, referral to otolaryngology may be delayed or deferred. In our study, patients with HLHS had significantly shorter median follow-up time which may account for their poor outcome with respect to vocal fold function, because they may have been more likely to follow-up before a natural return of vocal fold movement.

Aerodigestive Interventions

We noted a high incidence (46%) of surgical feeding tube placement in these children. This is likely multifactorial in nature. Vocal fold immobility certainly predisposes to aspiration and placement of a feeding tube may be undertaken as a protective measure. In a systematic review, Strychowsky et al identified an odds ratio of 7.3 (95% CI: 1.6, 32.8) for gastrostomy tube usage at the time of discharge in children who had a known vocal fold immobility after cardiothoracic surgery.[3] Sachdeva et al also found in their study of 36 infants with vocal fold immobility after cardiovascular surgery that 55.2% were on a tube feed only regimen at discharge.[4] Skinner et al reported that 48% of neonates undergoing the Norwood procedure had abnormalities on a modified barium swallow study post-operatively, with 24% showing frank aspiration.[11] However, Pereira et al reported that out of seven infants with left vocal fold immobility after PDA ligation, none required placement of a supplemental feeding tube.[13] The contrast between Sachdeva's and Skinner's results compared to Pereira's may be because the first two studies examined infants and neonates undergoing correction of large structural defects with much longer operative times and more extensive dissection near the recurrent laryngeal nerve. In Pereira's study, 5/7 infants had adequate compensation by the contralateral vocal fold whereas the contralateral vocal fold status was not specifically addressed in the two other studies.[13] It is possible that the contralateral vocal fold may not be compensating as well in children who have undergone more significant procedures and this may leave them at higher risk for requiring tube feeds. This could either be due to more extensive surgical dissection putting both recurrent laryngeal nerves at risk, longer post-operative intubation affecting laryngeal and pharyngeal function or general deconditioning. Prolonged and traumatic intubation and congenital immobility are also possible in these patients. These patients do not routinely have pre-operative FOEs to diagnose pre-operative vocal fold immobility.

CONCLUSION

In conclusion, children who have congenital heart disease requiring aortic arch manipulation and/or reconstruction are at risk of persistent vocal fold immobility after their surgical repair. These children require prolonged post-operative monitoring by otolaryngology and speech pathology to verify that they regain vocal fold function and there are no persistent voice or swallowing deficits. Parents and primary care providers should be counseled that their child has an approximately 46% chance of requiring a surgical feeding tube if a vocal fold immobility occurs after cardiovascular surgery, especially in HLHS. Once a surgical feeding tube is placed, the majority retain the tube for several years. Additionally, there is a high likelihood of requiring further operative airway interventions such as laryngoscopy/bronchoscopy or vocal fold medializations along with the need for feeding and speech therapy. They should also be counseled that if the vocal fold movement returns, it will likely be within one year, but in some cases recovery may still occur later.

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CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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