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Outcomes of recurrent laryngeal nerve injury following congenital heart surgery: A contemporary experience



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Objective: Injury to the recurrent laryngeal nerve can lead to significant morbidity during congenital cardiac surgery. The objective is to expand on the limited understanding of the severity and recovery of this iatrogenic condition.

Design: A six-year retrospective review of all congenital heart operations at a single institution from January 1, 2008 to December 31, 2013 was performed. All patients with documented vocal cord paralysis on laryngoscopic examination comprised the study cohort. Evaluation of time to vocal cord recovery and need for further surgical intervention was the primary focus.

Results: The incidence of post-operative vocal cord paralysis was 1.1% (32 out of 3036 patients; 95% confidence interval: 0.7–1.5%). The majority were left-sided injuries (71%). Overall rate of recovery was 61% with a median time of 10 months in those who recovered, and a total follow up of 46 months. Due to feeding complications, 45% of patients required gastrostomy tube after the injury, and these patients were found to have longer duration of post-operative days of intubation (median 10 vs. 5 days, $p = 0.03$), ICU length of stay (50 vs. 8 days, $p = 0.002$), and hospital length of stay (92 vs. 41 days, $p = 0.01$). No pre-operative variables were identified as predictive of recovery or need for gastrostomy placement.

Conclusion: Recurrent laryngeal nerve injury is a serious complication of congenital heart surgery that impacts post-operative morbidity, in some cases leading to a need for further intervention, in particular, gastrostomy tube placement. A prospective, multi-center study is needed to fully evaluate factors that influence severity and time to recovery.

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Introduction

Congenital heart defects are the most common malformation found in newborns [1,2], of which nearly a third require surgical repair [1]. Post-operative vocal cord dysfunction is a known complication of congenital heart surgery (CHS), and is often associated with several respiratory and gastrointestinal complications that can be severe to the point of necessitating further surgical intervention [3]. Reported incidence of vocal cord paralysis (VCP) following cardiac surgery ranges widely from 1.7% to 67% [4]; however, most studies indicate the rate is less than 9%. This difference is largely attributed to whether routine clinical laryngoscopic exam is performed, which increases the rate of diagnosis compared with examination of only symptomatic patients. The two most common mechanisms implicated involve direct trauma either to the recurrent laryngeal nerve (RLN) during the operation or indirect injury to the nerve from intubation [5]. Following CHS, it has been a challenge to discern which mechanism is responsible. The left RLN is at higher risk for VCP than the right side, particularly during aortic arch procedures, due to its proximity to the surgical plane of dissection [4]. Many post-operative cases resolve over time with supportive care. In persistent cases of VCP, external laryngoplasty implants as well as injection laryngoplasty have both provided durable improvement of symptoms [6,7].

Patients with the injury typically present with stridor, hoarseness, and a breathy and weak cry, while severe cases result in aspiration and respiratory distress. Reports of recovery rates from VCP following cardiac surgery range widely from 0% to 82%. The lowest recovery rates have been found in patent ductus arteriosus (PDA) ligations; the highest after thyroid or purely cardiac operations [8]. Few studies have been published regarding the clinical outcomes of this population, and therefore there is limited knowledge of outcomes and predictors of injury resolution. The aim of this study is to review the injury characteristics, mode of presentation and outcomes of patients with vocal cord paralysis in a high volume CHS center.

Methods

From January 1, 2008 through to December 31, 2013, a total of 3036 cases of CHS were performed at the Children's National Medical Center (CNMC) in Washington, DC. The CNMC

Abbreviations

CHS	congenital heart surgery
GERD	gastro-esophageal reflux disease
ICU	intensive care unit
MBS	modified barium swallow
PDA	patent ductus arteriosus
RLN	recurrent laryngeal nerve
VCP	vocal cord paralysis

Institutional Review Board approved the retrospective review and informed consent was waived. All patients presenting with postoperative stridor or any other clinical signs of VCP underwent either flexible fiberoptic laryngoscopy or rigid laryngobronchoscopic examination by a pediatric otolaryngologist. Gastrostomy tube placement was performed in all patients with feeding difficulty resulting in poor weight gain as a direct or indirect consequence of VCP, patients with evidence of aspiration on barium swallow, and patients with severe gastroesophageal reflux.

A retrospective chart review was carried out, inclusive of all patients aged 18 years or below with confirmed new onset VCP documented on laryngoscopic exam following a cardiac or aortic operation. Pre-operative, intra-operative and post-operative variables were evaluated. Univariate analysis was done using the nonparametric Mann-Whitney U-test for continuous variables and Fisher's exact test for binary proportions. Wilson's method was used to derive 95% confidence intervals around the observed incidence of RLN injury and recovery [9]. Time to recovery was based on documentation of cord mobility on laryngoscopic examination or resolution of symptoms when repeat laryngoscopy was not performed. Kaplan-Meier time-to-event analysis was used for estimating recovery of VCP during the follow-up period with 95% confidence intervals around the curve constructed using Greenwood's formula [10]. Statistical analysis was performed using IBM SPSS Statistics (version 21.0, IBM, Armonk, NY). Two-tailed $p < 0.05$ were considered statistically significant.

Results

Thirty-two patients over the six-year period were identified as having post-operative VCP, representing 1.1% of those undergoing CHS. One patient was excluded from the analysis due to inadequate follow up and incomplete data set. Of the remaining 31 injuries, 16% had bilateral VCP, 13% had right-sided VCP, and 71% had

left-sided VCP. Patients had undergone a range of cardiac procedures including 23 PDA ligations, 29 aortic arch reconstructions, five Norwood stage 1 cases as well as several types of intra-cardiac repairs and shunts (four ventricular septal defect repair, four atrial septal defect repairs, two Blalock–Taussig shunts, five aortic arch reconstructions, two Sano shunts, and one total anomalous pulmonary venous reconstruction). The age range of patients was 7–41 days (Table 1). The most common presenting symptom was stridor (74%), followed by hoarseness (29%) and weak cry (25.8%) (Table 1).

Sixty-one percent had post-operative feeding difficulties including aspiration, oropharyngeal dysphagia, or both (Table 3). Forty-five percent had documented aspiration or pharyngeal dysphagia, which was documented on modified barium swallow. Interestingly, surgical gastrostomy tube was required prior to discharge in 45% of patients with VCP, while another 13% were discharged with nasogastric tube only. Gastroesophageal reflux disease was diagnosed post-operatively in 68% of patients. Concomitant laryngeal pathologies, such as vocal cord granulomas, subglottic stenosis, tracheomalacia and bronchomalacia were discovered in 26% of patients, and central nervous system comorbidities were diagnosed in 16%. Additional airway operations were required in 23% of patients. Three patients (10%) necessitated tracheostomy placement, two of whom had bilateral VCP.

When comparing patients requiring gastrostomy tube placement to those who did not, significantly more patients had longer post-operative intubation times (median 10 vs. 5 days, $p = 0.03$), longer post-operative time to diagnosis of VCP

Table 2. Patient outcomes, comorbidities and procedures (based on cohort of 31 patients).

Persistent airway symptoms	12 (39%)
Another airway procedure	7 (23%)
G-tube	14 (45%)
CNS comorbidity	5 (16%)
Laryngeal pathology	8 (26%)
Postoperative GERD	21 (68%)
MBS aspiration	14 (45%)
Postoperative feeding difficulties	19 (61%)
Tracheostomy	3 (10%)
Mortality	2 (6.5%): 1 and 3 years follow up

MBS = modified barium swallow; GERD = gastroesophageal reflux disease.

(median 43 vs. 19 days, $p = 0.04$), longer ICU length of stay (median 50 vs. 8 days, $p = 0.002$), and longer hospital length of stay (median 92 vs. 41 days, $p = 0.01$) (Table 2).

The rate of recovery from VCP in our cohort was 61% ($n = 19$), while the remaining 39% had persistent airway symptoms at last follow up. Median follow-up time was 385 days (ranging from one month to three years) in patients who did not recover. Patients with and without recovery did not differ significantly in pre-operative variables, as shown in Table 1. Median time from cardiac surgery to diagnosis of VCP was 11 days shorter for patients with resolution (27 compared to 38 days) but the difference was not significant ($p = 0.8$). Two patients died: one related to heart failure and central nervous insult in the setting of pulmonary hypertensive crisis; the other from post-operative complications involving septic shock.

Median time to recovery was 10 months, ranging from one to 48 months. Kaplan–Meier analysis

Table 1. Comparison of patients with and without resolution of vocal cord paralysis.

Variable	Resolution of VCP ($n = 19$)	No Resolution of VCP ($n = 12$)	p value
Gender (M/F)	14/5	6/6	0.26
Age at surgery, d	21 (7–41)	11 (6–24)	0.41
Weight, kg	2.6 (1.2–3.6)	2.7 (1.1–3.6)	0.98
Prematurity	10 (53%)	5 (42%)	0.72
Extracardiac genetic	2 (11%)	1 (8%)	1.00
Aortic operation	17 (90%)	12 (100%)	0.51
PDA ligation	14 (74%)	9 (75%)	1.00
Norwood stage 1	3 (16%)	2 (17%)	1.00
Median sternotomy	2/10 (20%)	4/8 (50%)	0.32
ICU re-admission	1 (5%)	0 (0%)	1.00
Bilateral RLN injury	1 (5%)	4 (33%)	0.06
Stridor	13 (68%)	10 (83%)	0.43
Postop. time to dx , d	27 (16–102)	38 (18–87)	0.80
Initial ICU stay, d	16 (6–53)	29 (7–93)	0.44
Hospital stay, d	51 (33–108)	45 (31–98)	0.89

VCP = vocal cord paralysis; RLN = recurrent laryngeal nerve. Continuous variables including age, weight, time to diagnosis, ICU stay, and hospital length of stay are median (interquartile range).

estimated that 65% of patients will recover within 24 months of follow up (95% CI 50–80%) (Fig. 1).

Discussion

Injury to the RLN can be a severe complication of cardiac surgery because it leads to airway compromise by VCP, and also causes aspiration from loss of sensory and motor function to the cervical esophagus and pharynx. VCP symptoms include dysphonia, vocal fatigue, inefficient cough, or more concerning signs of respiratory distress including stridor. The incidence of VCP in our study was 1.1%, which corresponds to the lower end of the range of previous reports [4]. In our institution we have shifted our practice to a compulsory evaluation of every case of post-operative stridor by a pediatric otolaryngologist, even without accompanying clinical signs of feeding intolerance, aspiration, or increased supplemental oxygen requirement. The presence of stridor, hoarseness, weak cry, or signs of respiratory distress at any time following initial post-operative extubation prompts evaluation by flexible fiberoptic laryngoscopy. The decision to opt for rigid laryngobronchoscopy was made at the discretion of the otolaryngologist.

The operations were performed by one of four attending surgeons at our institution, each of whom uses the same intra-operative protocol for recurrent laryngeal nerve protection. The standard technique for avoiding nerve injury is to identify and gently dissect it away from the operating field during PDA ligations and cases involving the aortic arch. We include the use of intracardiac vascular catheters rather than

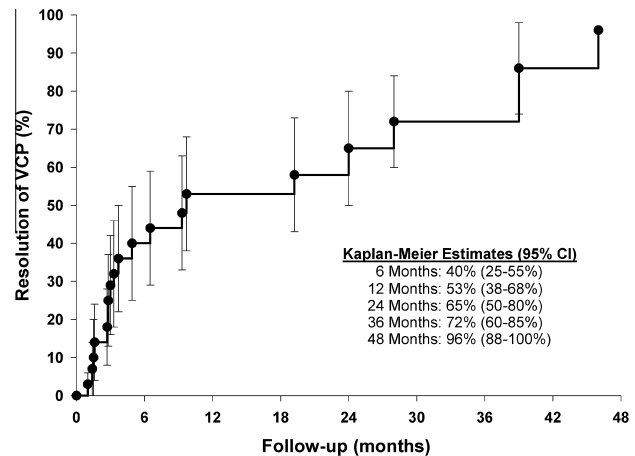


Figure 1. Time to recovery. Kaplan–Meier analysis of time to recovery shows 65% of patients with resolution of VCP had recovered by 24 months (95% CI 50–80%).

percutaneous, avoid ice slush, maintain low power protected-tip diathermy at 6–8 watts, and minimize use of retractors when dissecting out the aortic arch.

A study aiming to find predictors of RLN injury during cardiac surgery used a laryngeal sensor during 15 cases of mediastinoscopy and inferred that RLN injury was most readily caused by traction during paratracheal dissection [12]. Notably higher rates of VCP have been found during patent ductus arteriosus ligations, which are attributed to the anatomic proximity [13].

A previous comprehensive review of RLN injuries suggested eight mechanisms for the injury, aside from direct surgical trauma: central venous catheterization, hyperextension of the neck, traumatic endotracheal intubation, endotracheal tube

Table 3. Comparison of patients with and without need for gastrostomy tube procedure.

Variable	G-tube (n = 14)	No G-tube (n = 17)	p value
Gender (M/F)	8/6	12/5	0.48
Age at surgery, d	9 (6–42)	22 (8–39)	0.42
Weight, kg	2.7 (1.3–3.6)	2.4 (1.1–3.7)	0.86
Prematurity	6 (43%)	9 (53%)	0.72
Extracardiac genetic	1 (7%)	2 (12%)	1.00
Aortic operation	12 (86%)	17 (100%)	0.20
PDA ligation	10 (71%)	13 (77%)	1.00
Norwood stage 1	3 (21%)	2 (12%)	0.64
Median sternotomy	3/8 (38%)	3/10 (30%)	1.00
ICU readmission	1 (7%)	0 (0%)	0.45
Bilateral RLN injury	3 (21%)	2 (12%)	0.64
Stridor	10 (71%)	13 (77%)	1.00
Postop. time to dx, d	43 (24–118)	19 (13–86)	0.04*
Postop. intubation, d	10 (5–22)	5 (3–11)	0.03*
Initial ICU stay, d	50 (16–113)	8 (4–31)	0.002*
Hospital stay, d	92 (42–133)	41 (11–56)	0.01*

Age, weight, time to diagnosis, ICU stay, and hospital length of stay are median (interquartile range).

* Statistically significant.

trauma, faulty insertion of the nasogastric tube, sternotomy, traction on the heart, and hypothermic injury from ice slush [11]. As mentioned previously, our team takes measures to protect against several of these and other potential risk factors, which may explain why our rates are lower than previous reports.

Reported recovery rates have ranged from 0% to 82% [8], but few studies, if any, have previously commented on predictive factors of recovery from VCP. Our analysis shows 61% recovery after 48 months of follow up. No preoperative factors were found to be significantly predictive of poor recovery, although power is limited given the small sample size. Given our extreme caution to protect the nerve intra-operatively, we hypothesize that many RLN injuries are not due to laceration, crush, or thermal injury but rather caused by inadvertent stretch. We therefore evaluated factors that may affect size and shape of the thoracic cavity and the anatomic relationships therein, including patient age, weight, prematurity, and gender as potential contributors to stretch injury. Likewise, the specific procedures undertaken theoretically have consistently disparate risk of nerve injury and thus were also explored.

Days of postoperative intubation as well as the attendant prolonged ICU and hospital stays were risk factors for requirement of further operations in the form of gastrostomy tube placement. This relates to prolonged pharyngeal and vocal cord trauma from the endotracheal tube; however, it is unclear whether the prolonged intubation in these cases was due to an operation involving greater trauma to the RLN. Furthermore, it is unknown whether VCP is more pronounced from direct trauma to the cords as opposed to RLN traction. The findings from our patients confirm previous studies showing increased rates of gastrostomy tube requirement for pharyngeal dysfunction up to 67% in patients suffering from VCP [14]. Our study demonstrates 45% of these injuries required gastrostomy, and another 14% were discharged with nasogastric tubes. Although predictors of need for gastrostomy were not identified in our study, it is interesting that in clinical practice these are patients who tend to have other associated findings such as aspiration and gastroesophageal reflux, and further exploration of these factors is necessary.

An inherent limitation of the current study is the small sample size, which reduced statistical power for detecting predictors of recovery. Indeed, greater statistical power could reveal risk factors for recovery as well as gastrostomy tube

placement that could be managed more effectively preoperatively or in the operating room. Further, the retrospective nature of this study may underestimate the true incidence and recovery rate, particularly in those patients who had VCP but were asymptomatic. This review highlights the advanced morbidity associated with VCP after congenital cardiac surgery. To the best of our knowledge, no other study has evaluated potential predictors of recovery. A multicenter prospective review of a large cohort is needed for adequate power to identify independent predictors of injury severity and factors that potentially influence time to recovery. Such factors could guide clinical management to limit severity and recovery time of VCP.

Author contributions

Concept/design: F.A., C.H., G.A., R.C., M.R., C.S., B.R., D.Z., R.J., D.N.; Data analysis/interpretation: F.A., C.H., D.Z., D.N.; Drafting article: F.A., C.H., D.Z., D.N.; Critical revision of article: F.A., C.H., B.R., D.Z., R.J., D.N.; Approval of article: F.A., C.H., G.A., R.C., M.R., C.S., B.R., D.Z., R.J., D.N.

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