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Outcomes of pediatric tracheostomy after surgery for congenital heart disease: A 20-year experience

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Abstract

Objective: Children with congenital heart defects (CHD) requiring cardiovascular surgery (CVS) rarely require tracheostomy placement; however the mortality rate remains high. The study aimed to analyze the incidence of tracheostomy in children with CHD, and to determine factors contributing to postoperative outcomes, decannulation rates, and mortality.

Methods: Retrospective case series of children ≤18 years old with CHD status post-CVS who underwent tracheostomy placement between January 1, 2001 and December 31, 2020. Variables analyzed included demographic information, presence of comorbidities including prematurity, respiratory diseases, presence of genetic syndromes, decannulation status, type of repair (univentricular vs. biventricular), and need for cardiopulmonary bypass. Adverse events analyzed included all-cause mortality, development of mediastinitis, fatal decannulation, and persistence of tracheocutaneous fistula.

Results: Fifty-one patients were analyzed. The incidence of tracheostomy was 0.8%. Median age at tracheostomy was 5.3 months. The 5-year survival estimate was 56.3% (95% confidence interval 43.6%, 72.6%). Age ≤6 months at the time of tracheostomy placement ($p = .03$), and the presence of tracheomalacia ($p = .04$) were factors significantly associated with 5-year survival. Two patients (3.9%) experienced fatal decannulation, and one patient (2.0%) developed postoperative mediastinitis. The 10-year decannulation rate estimate was 47.8% (30.5%, 63.2%). Seven patients (13.7%) had a persistent tracheocutaneous fistula.

Conclusions: This study corroborates high mortality rates in this population. Factors associated with improved survival were younger age at the time of tracheostomy and presence of tracheomalacia. Decannulation rates were low, but estimates improved over 10 years. Further studies are needed to determine optimal indications and timing for tracheostomy placement in this patient population.

Level of Evidence: 4

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KEYWORDS
congenital heart disease, mortality, pediatric tracheostomy, tracheostomy-related complications

1 | INTRODUCTION

Children with congenital heart defects (CHD) rarely require tracheostomy placement, with an estimated incidence of 0.8%–2.7%; however, evidence has shown high mortality rates in this patient population.[1–6] The incidence of tracheostomies performed on children with CHD has increased over the last two decades, and this may be attributed to improved survival of children with CHD, who might not have survived in the past without current medical and surgical advances.[4] Despite this, mortality rates in children with CHD who undergo tracheostomy remain high and can range from 23.5% to 52.4%.[2,3,5,7] Preoperative risk factors associated with tracheostomy placement in these children include underlying chromosomal abnormalities, prematurity, younger age, low birth weight (<1000 g), bronchodysplasia, and underlying pulmonary disease.[6] In addition, multiple factors have been associated with increased mortality in this population, including prolonged time to tracheostomy, pulmonary comorbidities, and univentricular physiology.[3,8,9]

Support for early tracheostomy placement relates to decreased need for sedation, potential for more rapid wean from ventilator support and improved hemodynamics, shorter intensive care unit stays, and improved pulmonary toilet. Placing a tracheostomy in children who have undergone, or who will be undergoing, surgery for management of their CHD raises concern for development of mediastinitis given proximity of the tracheostoma to the median sternotomy incision. There is relatively limited data regarding the impact of tracheostomy placement relative to the incidence of mediastinitis in those with CHD.[5,6,10] Furthermore, there is limited data on long-term survival and decannulation rates in this population. Overall health status of the patient, preference of the cardiovascular surgeon, and parental decision to move forward with tracheostomy all likely play a role in the timing of the procedure.

There has been a perceived increase in acuity and need for tracheostomy placement in children with CHD undergoing cardiovascular surgery (CVS) at our institution. Given this, the goals of this study were to characterize the patient population undergoing tracheostomy after surgery for CHD, with aims toward a better understanding of the mortality rates, and the potential decannulation rates. Additionally, the analysis will seek to identify risk factors for mortality, particularly those suggestive of a recommendation for earlier tracheostomy placement.

2 | MATERIALS AND METHODS

This study was approved by the Institutional Review Board of Ann & Robert H. Lurie Children's Hospital of Chicago (IRB no. 2021–5037). This is a retrospective case series of children under the age of 18 with CHD, who underwent tracheostomy placement by the otolaryngology service at our institution between January 1, 2001 and December 31, 2020. Data were collected from an active database of CVS patients maintained by the cardiovascular service, and from the electronic health record (EHR). Children who had their tracheostomy placed at an outside institution were excluded. Children who had non-cardiac thoracic procedures (i.e., laryngotracheal reconstruction procedures), or who had tracheostomy placement prior to CVS, were also excluded from the analysis.

Variables analyzed included sex, race, ethnicity, prematurity, age at tracheostomy, time from cardiac surgery to tracheostomy placement, duration of intubation prior to tracheostomy, type of surgical repair (univentricular or biventricular), need for cardiopulmonary bypass, presence of genetic syndromes, and airway comorbidities including tracheomalacia, pulmonary hypertension, and bronchopulmonary dysplasia (BPD). Outcomes after tracheostomy included all-cause mortality, time to decannulation, and adverse outcomes including mediastinitis, fatal decannulation, and persistence of a tracheocutaneous fistula. For patients with missing data, data were reported as "Unknown," and these patients were excluded from the pertinent univariate analysis.

2.1 | Statistical analysis

Continuous variables were reported as median (interquartile range), whereas categorical variables were reported as percentages. Kaplan–Meier estimates were used to compute 1- and 10-year overall survival rates after tracheostomy placement and were reported as rate (95% confidence interval [CI]). Rates of tracheostomy decannulation were adjusted for the competing risk of death and reported as the cumulative incidence.[11] The log-rank test was used to study the association of multiple parameters in relationship to 10-year all-cause mortality (sex, race, ethnicity, prematurity, age at tracheostomy, time from cardiac surgery to tracheostomy placement, duration of intubation prior to tracheostomy, type of cardiac surgical repair, need for cardiopulmonary bypass, presence of genetic syndromes, presence of tracheomalacia, pulmonary hypertension, and BPD). Significance level was set as $\alpha = .05$. All statistical analyses were performed using R version 4.2.2 (R Foundation for Statistical Computing, Vienna, Austria).

3 | RESULTS

From January 2001 to December 2020, CVS was performed on a total of 6359 patients. Fifty-one of these patients underwent postoperative tracheostomy placement. The overall incidence of tracheostomy placement in this population over the study period was 0.8%.

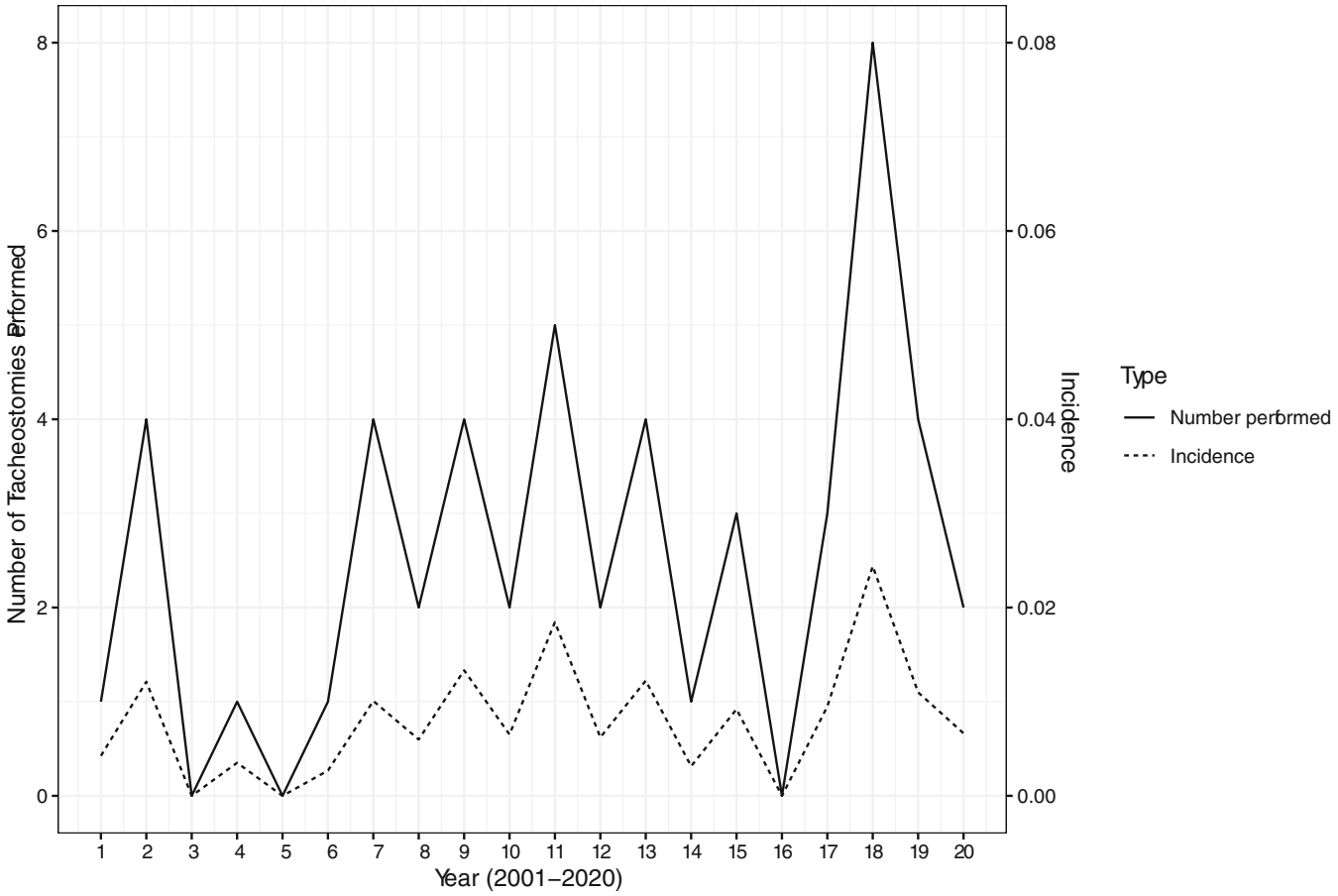

FIGURE 1 Incidence of tracheostomy placement in children after surgery for congenital heart disease.

The incidence over the 20-year time period of the analysis is summarized in Figure 1.

Characteristics of the 51 patients identified in our study are summarized in Tables 1 and 2. Of those who underwent tracheostomy, 26 (51.0%) were male, 26 (51.0%) were white, 16 (31.4%) were black or African American, and 6 (11.8%) were of Hispanic ethnicity. The median age at the time of tracheostomy was 5.3 months (2.8–7.4 months). A total of five (9.8%) patients had Trisomy 21, seven (13.7%) 22q deletion syndrome, three (5.9%) Turner syndrome, and seven patients had other syndromes (13.7%). Twenty (39.2%) patients had BPD, 10 (19.6%) pulmonary hypertension, and 35 (68.6%) tracheomalacia. The median time between cardiac surgery and tracheostomy was 57 days (38.5–101.5). Prior to tracheostomy, seven patients (13.7%) did not require intubation and one patient's length of intubation was unknown. Five of these seven patients (71.4%) underwent tracheostomy placement due to inability to wean respiratory support from high-flow nasal cannula or noninvasive positive pressure ventilation. The indications for the remaining two patients were severe obstructive sleep apnea and bilateral vocal fold paresis, respectively. For those who did require intubation, the median length of intubation prior to tracheostomy was 54 days (24.5–92.5 days). Univentricular repair was performed in 12 (23.5%) patients, and cardiopulmonary bypass was used in 39 (76.5%) cases.

Median follow-up time was 2.6 years (0.45–7.98 years). A total of 37 patients (72.5%) patients survived to hospital discharge. Table 3 demonstrates overall survival rates after tracheostomy placement stratified by baseline and perioperative characteristics. Overall survival (95% CI) was estimated as 68.1% (56.4%, 82.3%) at 1 year, 61.5% (49.3%, 76.8%) at 3 years, and 56.3% (43.6%, 72.6%) at 5 years. Two patients (3.9%) experienced a fatal decannulation event at home, 2.0 and 3.2 years postoperatively. One patient (2.0%) experienced postoperative mediastinitis. The patient was placed on a 42-day course of meropenem for pseudomonal mediastinitis and received gentamicin washes to the open chest area. The patient was briefly decannulated, and the tracheostomy was later replaced due to lack of spontaneous respiratory effort. Care was withdrawn 5 months postoperatively.

Rates of tracheostomy decannulation (Figure 2), adjusted for the competing risk of death, were estimated as 4.1% (0.7%, 12.5%) at 1 year, 20.0% (9.6%, 33.1%) at 3 years, 36.6% (21.9%, 51.4%) at 5 years, and 47.8% (30.5%, 63.2%) at 10 years. Seven patients (13.7%) had a persistent tracheocutaneous fistula after decannulation. The diagnosis of tracheomalacia ($p = .04$) and age under 6 months at the time of tracheostomy ($p = .03$) were identified as significant factors associated with improved 5-year survival.

TABLE 1 Patient baseline characteristics.

Patient characteristic ($n = 51$)	No. or median	% or IQR
Sex, male	26	51.0%
Age at tracheostomy surgery (months)	5.3	2.8–7.4
Race		
White	26	51.0%
Black or African American	16	31.4%
Other	7	13.7%
Unknown	2	3.9%
Ethnicity		
Hispanic	6	11.8%
Non-Hispanic	42	82.4%
Unknown	3	5.9%
Comorbidities		
Prematurity	24	47.1%
22q deletion syndrome	7	13.7%
Trisomy 21	5	9.8%
CHARGE syndrome	4	7.8%
Turner syndrome	3	5.9%
Other syndrome	7	13.7%
Bronchopulmonary dysplasia	20	39.2%
Pulmonary hypertension	10	19.6%
Tracheomalacia	35	68.6%
Length of follow-up (years)	2.6	0.45–7.98

Abbreviation: IQR, interquartile range.

TABLE 2 Perioperative details and adverse events.

Patient characteristic ($n = 51$)	No. or Median	% or IQR
Univentricular cardiac repair	12	23.5%
Cardiopulmonary bypass used	39	76.5%
Time between cardiac surgery and tracheostomy (days)	57	38.5–101.5
Intubated prior to tracheostomy	43	84.3%
Days intubated prior to tracheostomy[a]	54	24.5–92.5
Adverse events		
Death prior to hospital discharge	14	27.5%
Persistent tracheocutaneous fistula	7	13.7%
Fatal decannulation event	2	3.9%
Postoperative mediastinitis	1	2.0%

Abbreviation: IQR, interquartile range.
[a]One patient with unknown length of intubation and seven patients who did not require intubation were excluded.

4 | DISCUSSION

This was one of the largest single-center studies of tracheostomy outcomes in pediatric patients who have undergone surgery for congenital heart disease and included 20 years of clinical data. The analysis described the patient clinical characteristics, incidence of tracheostomy placement, mortality rates, decannulation rates, and identified clinical factors associated with improved 5-year overall survival. The overall incidence of tracheostomy placement was 0.8%, and the 1-year survival rate estimate was between 56.4%, 82.3%, consistent with prior literature.[2,3,5,7] The analysis described outcomes at 5 and 10 years, for which limited data exists.[2,9] Estimates of 5-year survival range from ~50% to 70%,[2,9] similar to the findings in our study (43.6%, 72.6%). Tracheostomy decannulation rate estimates were also described and indicated that decannulation rates were low for several years after tracheostomy placement, but they approached 50% for this population at 10 years. These long-term outcomes could be informative to families as they make decisions regarding tracheostomy placement.

Although the rate of fatal decannulation events in the study population (3.9%) was lower than reported in the literature,[2] this remains an important target for new and ongoing quality and safety initiatives. Placement of tracheostomy in CVS patients also raised concern for the development of mediastinitis given the proximity of the tracheostoma to the median sternotomy incision; however, this outcome was relatively rare in our study, occurring in only one patient (2.0%). This rate was lower than a previous report.[5] These rates may represent underestimates due to our small sample size, and the limited availability of postdischarge event data in the EHR.

In this study, univariate analysis revealed children who were younger at the time of tracheostomy placement, and those who had tracheomalacia, had better survival rates. The relationship between younger age and survival is in opposition to existing data regarding the broader population of patients with tracheostomies. A retrospective single-center study by Funamura et al.[4] identified 513 children (20% with CHD) who underwent tracheostomy, and they found a significant association between older age at the time of tracheostomy placement (13–18 years) and improved survival. Similarly, a prospective single-center study by Teplitzky et al.[12] identified 271 patients (45% with CHD), and they found a significant association between older age and improved survival. The age impact on mortality was not observed in the postoperative CHD population in two of the largest database studies by Mastropietro et al.[5] and Johnson et al.[3] The latter study did find a significant association between delayed timing of tracheostomy and decreased survival. Although our study did not mirror these findings, both findings in conjunction may serve as an impetus to consider earlier tracheostomy in select patients.

Our analysis identified an association between tracheomalacia and improved survival. Airway malacia is common in postoperative CHD patients, with estimates ranging from 23% to 66%.[7,9,10] The results of this study supported the finding that airway anomalies were associated with improved survival, as described by Johnson et al.[3] A plausible explanation for this phenomenon pertains to the transient nature of airway malacia, which may improve or resolve within the first few years of life.[13]

Although the type of cardiac repair has been demonstrated as an associated factor for increased mortality,[8] this finding did not

TABLE 3 Overall survival rate estimates after tracheostomy placement stratified by baseline and perioperative characteristics in patients who underwent tracheostomy after surgery for congenital heart disease.

	1-Year survival estimate (95% CI)	3-Year survival estimate (95% CI)	5-Year survival estimate (95% CI)	*p*-Value[a]
Overall	68.1% (56.4%, 82.3%)	61.5% (49.3%, 76.8%)	56.3% (43.6%, 72.6%)	
Sex				.28
Male	76.3% (61.3%, 94.8%)	67.7% (51.5%, 89.0%)	62.9% (46.1%, 85.7%)	
Female	60.0% (43.6%, 82.6%)	55.4% (38.8%, 79.1%)	49.2% (32.2%, 75.3%)	
Age at tracheostomy				**.03***
> 6 months	55.7% (37.7%, 82.2%)	45.0% (27.5%, 73.4%)	39.4% (22.6%, 68.6%)	
≤ 6 months	76.7% (62.9%, 93.4%)	72.8% (58.4%, 90.9%)	68.3% (52.9%, 88.1%)	
Race				.57
White	72.4% (56.9%, 92.1%)	72.4% (56.9%, 92.1%)	67.6% (51.2%, 89.1%)	
Black or African American	68.8% (49.4%, 95.7%)	61.9% (41.9%, 91.4%)	53.0% (32.4%, 86.9%)	
Other	71.4% (44.7%, 100.0%)	38.1% (13.7%, 100.0%)	38.1% (13.7%, 100.0%)	
Hispanic ethnicity				.25
Yes	66.7% (37.9%, 100.0%)	33.3% (10.8%, 100.0%)	33.3% (10.8%, 100.0%)	
No	71.0% (58.4%, 86.3%)	68.4% (55.6%, 84.2%)	61.7% (48.0%, 79.4%)	
Prematurity				.41
Yes	62.5% (45.8%, 85.2%)	57.3% (40.2%, 81.6%)	51.6% (34.2%, 77.7%)	
No	75.1% (59.7%, 94.5%)	66.5% (50.0%, 88.5%)	61.0% (43.7%, 85.0%)	
Genetic syndrome				.47
Yes	80.1% (65.9%, 97.4%)	67.0% (50.5%, 88.8%)	57.0% (39.8%, 81.7%)	
No	56.0% (39.6%, 79.3%)	56.0% (39.6%, 79.3%)	56.0% (39.6%, 79.3%)	
Pulmonary hypertension				.18
Yes	50.0% (26.9%, 92.9%)	50.0% (26.9%, 92.9%)	33.3% (12.1%, 91.7%)	
No	72.6% (60.1%, 87.8%)	64.3% (50.9%, 81.3%)	61.3% (47.5%, 78.9%)	
Bronchopulmonary dysplasia				.87
Yes	70.0% (52.5%, 93.3%)	70.0% (52.5%, 93.3%)	57.3% (38.4%, 85.4%)	
No	66.9% (52.0%, 86.0%)	56.3% (40.9%, 77.4%)	56.3% (40.9%, 77.4%)	
Tracheomalacia				**.04***
Yes	76.6% (63.6%, 92.2%)	73.5% (60.1%, 90.0%)	65.1% (50.0%, 84.7%)	
No	50.0% (30.6%, 81.6%)	37.5% (19.9%, 70.6%)	37.5% (19.9%, 70.6%)	
Type of repair				.18
Univentricular	50.0% (28.4%, 88.0%)	40.0% (19.6%, 81.8%)	40.0% (19.6%, 81.8%)	
Biventricular	73.8% (61.1%, 89.2%)	68.4% (55.0%, 84.9%)	61.3% (47.1%, 79.9%)	
Cardiopulmonary bypass used				.42
Yes	66.0% (52.6%, 82.9%)	60.1% (46.3%, 78.2%)	52.8% (38.4%, 72.6%)	
No	75.0% (54.1%, 100.0%)	66.7% (44.7%, 99.5%)	66.7% (44.7%, 99.5%)	
Time between cardiac surgery and tracheostomy				.35
> 60 days	65.2% (48.4%, 87.9%)	54.5% (36.9%, 80.6%)	46.7% (28.5%, 76.6%)	
≤ 60 days	70.6% (55.3%, 90.0%)	66.7% (51.0%, 87.1%)	62.5% (46.5%, 84.0%)	
Days intubated prior to tracheostomy[b]				.25
> 60 days	55.0% (37.0%, 81.8%)	55.0% (37.0%, 81.8%)	42.8% (25.2%, 72.6%)	
≤ 60 days	73.9% (58.0%, 94.2%)	59.9% (42.6%, 84.3%)	59.9% (42.6%, 84.3%)	

Abbreviation: 95% CI, 95% confidence interval.

[a]Log-rank test.

[b]One patient with unknown length of intubation and seven patients who did not require intubation were excluded.

*p-Value < .05.

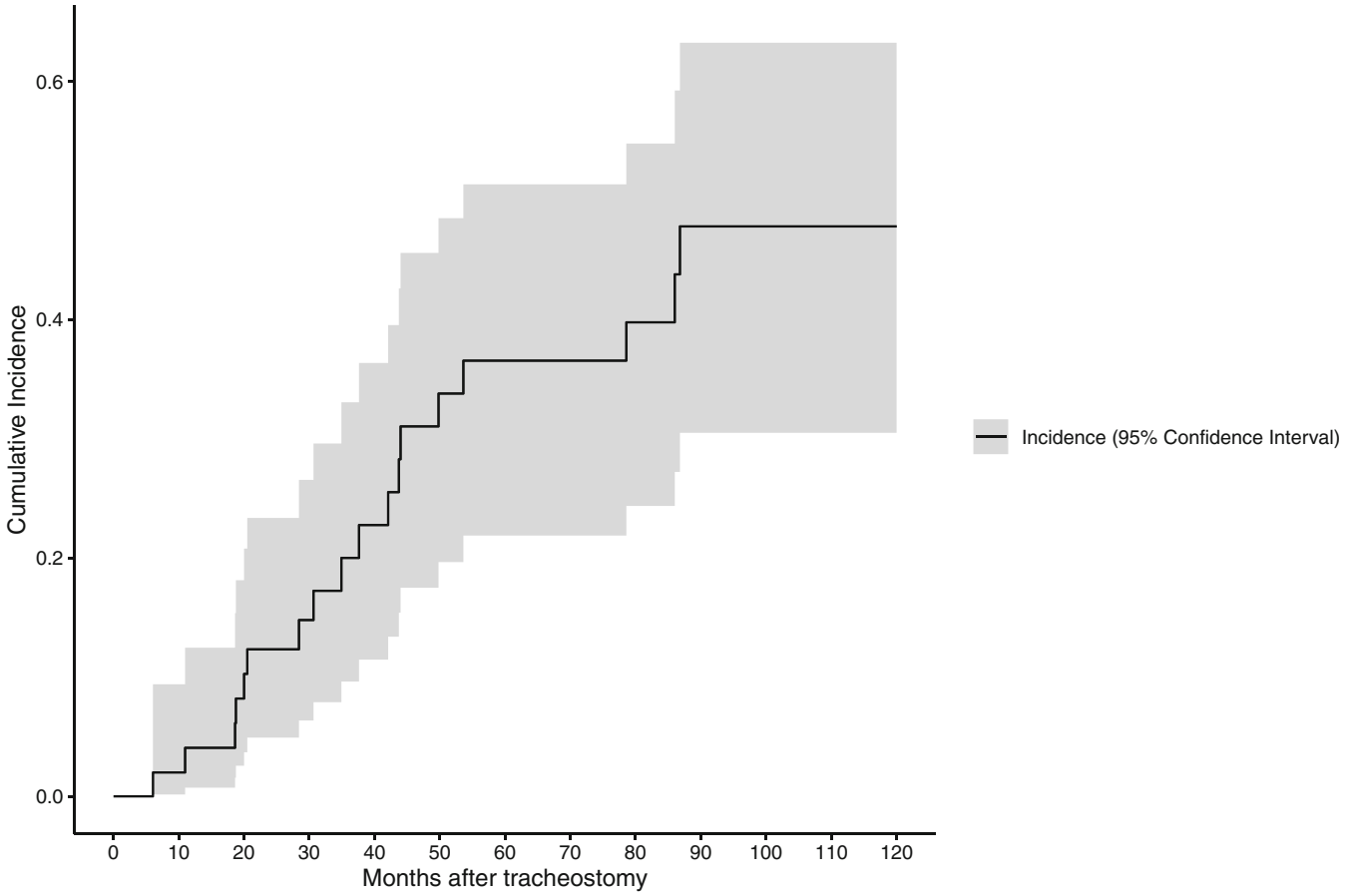

FIGURE 2 Cumulative 10-year decannulation estimates among pediatric patients with tracheostomy placement after surgery for congenital heart disease, adjusted for the competing risk of death. Estimates of decannulation rates (95% confidence intervals [CI]) were 4.1% (0.7%, 12.5%) at 1 year, 20.0% (9.6%, 33.1%) at 3 years, 36.6% (21.9%, 51.4%) at 5 years, and 47.8% (30.5%, 63.2%) at 10 years.

reach significance in the present study. A trend toward improved 5-year survival for those with biventricular physiology ($p = .18$) was observed. Increasing the number of patients analyzed, or performing multicenter analyses, may improve the ability to detect these differences.

Limitations of this study include the single center, retrospective nature of the design, and the limited patient population identified, increasing the study's susceptibility to selection bias, and potentially limiting generalizability. The study focuses on patients who undergo tracheostomy after CVS and did not consider patients with chronic cardiopulmonary failure who never underwent CVS prior to tracheostomy. The reported rates of adverse events may represent underestimates, especially when considering the limited availability of postdischarge event data in the EHR and potential loss to follow-up. Furthermore, there was a lack of consistent clinical protocols to guide decision-making around indications and timing of tracheostomy placement. There is heterogeneity within the population of CHD patients, and further analyses to characterize outcomes for patients with specific defects are needed. Nonetheless, this study contributed additional long-term data on mortality and decannulation rates after tracheostomy in postoperative CHD patients. Future directions include larger, prospective, multicenter studies, and development of care guidelines for this complex patient population.

5 | CONCLUSION

This study provided insights into tracheostomy outcomes in pediatric patients who had undergone surgery for CHD, including an overall low incidence of tracheostomy placement, and high mortality rates consistent with the literature. The analysis estimated long-term decannulation rates, which could be helpful when discussing outcomes with families. The study identified clinical factors associated with improved 5-year survival, including younger age at the time of tracheostomy placement, and the presence of tracheomalacia. Future research directions include larger, prospective, multicenter studies, and development of care guidelines for this complex patient population.

CONFLICT OF INTEREST STATEMENT

The authors have no disclosures or conflicts of interest.

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