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Indications and Outcomes of Tracheostomy in Children After Congenital Heart Surgery

Doğuştan Kalp Cerrahisi Sonrası Çocuklarda Trakeostomi Endikasyonları ve Sonuçları

Pınar Yazıcı Özkaya[1], Eşe Eda Turanlı[1], İrem Ersayoğlu[1], Osman Nuri Tuncer[2], Bülent Karapınar[1]

[1]Ege University Faculty of Medicine, Department of Pediatric Intensive Care, İzmir, Turkey
[2]Ege University Faculty of Medicine, Department of Cardiovascular Surgery, İzmir, Turkey

Abstract

Introduction: In a minority of children after cardiovascular surgery may require prolonged mechanical ventilatory support and tracheostomy. We aim to describe indications, timing and, outcomes of the tracheostomy.

Methods: A retrospective review of 12 children requiring tracheostomy after cardiac surgery between January 2010-December 2019 was performed. The patients' characteristics, indications and timing for tracheostomy, and survival were reviewed.

Results: After cardiac surgery, 12 (0.5%) of 2.459 patients with a median age at surgery of 210 days (interquartile range: 75-262 days) underwent tracheostomy. The median time between cardiac surgery and tracheostomy was 25 days (interquartile range: 15-47 days). Diaphragmatic paralysis was the most common (42%) indication for tracheostomy. Genetic syndrome or at least one non-cardiac morbidity was present in 41.6% of patients. The duration of mechanical ventilation was shorter in patients who had tracheostomy within 30 days compared with >30 days following intubation (30 vs. 60 days, p=0.035). The median length of pediatric intensive care unit stays after the tracheostomy was 41 days (range, 21-289 days). Among all patients with congenital heart surgery undergoing tracheostomy, 6 (50%) of 12 were decannulated after a median time of 179 days (range, 34-463 days). The operative mortality was 8.3% (1/12) and the overall mortality during the first year of follow-up was 8.3% (1/12).

Conclusion: An early tracheostomy procedure may facilitates the weaning process and shorten the duration of positive pressure ventilation.

Keywords: Tracheostomy, congenital heart surgery, prolonged mechanical ventilation, children

Öz

Giriş: Kardiyovasküler cerrahi sonrası çocukların çok az bir kısmında uzun süreli mekanik ventilasyon desteği ve trakeostomi gerekmektedir. Bu çalışma ile trakeostominin endikasyonlarını, zamanlamasını ve sonuçlarını tanımlamayı hedefliyoruz.

Yöntemler: Ocak 2010-Aralık 2019 tarihleri arasında kalp cerrahisi sonrası trakeostomi gerektiren hasta verileri geriye dönük olarak değerlendirildi. Hastaların preoperatif özellikleri, trakeostomi endikasyonları, trakeostomi açılma zamanı ve klinik sonuçları analiz edildi.

Bulgular: Kalp cerrahisi uygulanan 2,459 hastanın 12'sine (%0,5) trakeostomi açıldı. Bu hastaların kalp cerrahisi sırasında ortanca yaşı 210 gün (çeyrek değerler genişliği: 75-262 gün) idi. Kalp cerrahisi ile trakeostomi arasındaki ortalama süre 25 gündü (çeyrek değerler genişliği: 15-47 gün). Trakeostomi için en sık endikasyon postoperatif gelişen diyafragma paralizisi (%42) idi. Hastaların %41,6'sında genetik sendrom veya en az bir kardiyak olmayan morbidite mevcuttu. Mekanik ventilatörde kalış süresi değerlendirildiğinde, çocuk yoğun bakım yatışlarının ilk 30 gün içinde trakeostomi açılan hastaların mekanik ventilatörde kalma süresi, 30. gün sonrasında trakeostomi açılan hasta grubuna göre daha kısaydı (sırasıyla 30 gün, 60 gün, p=0,035). Trakeostomiden sonra çocuk yoğun bakım ünitesinde ortalama kalış süresi 41 gündü (aralık, 21-289 gün). Doğuştan kalp cerrahisi sonrası trakeostomi açılan 12 hastanın 6'sı (%50) ortalama 179 gün (aralık, 34-463 gün) sonra dekanüle edildi. İzlem sonrası birinci yılda operatif mortalite %8,3 (1/12) ve genel mortalite %8,3 (1/12) idi.

Sonuç: Doğuştan kalp cerrahisi sonrası trakeostomi ihtiyacı olan hastalarda erken trakeostomi işlemi, pozitif basınçlı ventilasyondan ayrılma sürecini kolaylaştırır ve pozitif basınçlı ventilasyon süresini kısaltabilir.

Anahtar Kelimeler: Trakeostomi, doğuştan kalp cerrahisi, uzamış mekanik ventilasyon, çocuk

Address for Correspondence/Yazışma Adresi: Pınar Yazıcı Özkaya, Ege University Faculty of Medicine, Department of Pediatric Intensive Care, İzmir, Turkey
E-mail: dryazicipinar@gmail.com **ORCID ID:** orcid.org/0000-0002-1209-2534
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Introduction

In the recent 20 years, improvement in the field of congenital heart surgery (CHS) and postoperative management result in improved survival in patients with congenital heart disease. Early extubation has advantages including reduced sedo-analgesia, decreased ventilator-associated pneumonia, and improved cardiovascular interactions. However, a small percentage of children who had CHS has a risk for prolonged ventilation. Prolonged mechanical ventilation is the most common indication for tracheostomy after CHS and occurs as a result of multiple etiologic factors. A large cohort of the patients who underwent tracheostomy after CHS reported that the incidence of tracheostomy increased from 0.11% to 0.76% between 2000-2012.[1]

Tracheostomy after cardiac operations appears to be associated with higher hospital mortality and higher mortality after discharge. Especially, children with single-ventricle physiology have lower long-term survival.[2] After CHS, in-hospital mortality following tracheostomy ranged from 7.7% to 28%.[3] However, the annual mortality rate of children with tracheostomy after CHS has not changed over time. It has reported 24.5% in 2010 and 27.5% in 2013.[1]

The purpose of the study is to describe the tracheostomy indications, complications, and long-term outcomes in children undergoing CHS.

Materials and Methods

We performed a retrospective chart review of all children who required tracheostomy after CHS from January 2010 to December 2019. Patients were identified from the pediatric intensive care unit (PICU) and cardiothoracic surgical database. All pediatric patients under the age of 14 years who underwent tracheostomy after CHS were included. Neonates and patients with tracheostomies placed before cardiac surgery were excluded. The demographic data, cardiac diagnosis, details of surgical procedures such as cross-clamp and cardiopulmonary bypass duration, and Risk Adjustment for Congenital Heart Surgery Score (RACHS)-1 were recorded.[4] Patients were evaluated for several extubation trials, tracheostomy timing, and duration of mechanical ventilation before and after the tracheostomy. We also recorded the Pediatric Risk of Mortality Score (PRISM)-III at PICU admission, which is a validated and physiology-based scoring system for rating the severity of medical illness for children.[5] The number of ventilator-associated pneumonia (VAP) episodes was recorded before and after tracheostomy. The indication for tracheostomy and, complications related to tracheostomy were recorded. This study was approved by the institutional review boards with the permission for the use of patient data for publication purposes (21-1T/26). Informed consent was received from the families.

Our primary outcomes included operative mortality and long-term survival, which was defined as 1-year survival. We reported operative mortality according to the definition of the Society of Thoracic Surgeons (STS) and Congenital Heart Surgery Database. Other outcomes include tracheostomy incidence, length of stay, time of decannulation if the patient had been decannulated, and mechanical ventilatory need after hospital discharge.

Statistical Analysis

Statistical analysis was performed using IBM SPSS Statistics version 20. Descriptive data are expressed as means (standard deviations) or medians (ranges) as appropriate. Categorical data were compared using the chi-squared test or Fisher's Exact test. Continuous data were analyzed using the Mann-Whitney U test for non-normally distributed data.

Results

A total of 12 patients underwent tracheostomy placement following cardiac surgery in the 9 year study period. During this period 2459 CHS performed in our center, the incidence of tracheostomy after CHS was 0.5%. Eight of the 12 patients (66.7%) were female. The median weight at the time of surgery was 6 kg (range, 4 kg-35 kg).

The median age at the time of surgery was 210 days [interquartile range (IQR): 75-262 days]. The median time between cardiac surgery and tracheostomy was 25 days (IQR: 15-47 days). The mean PRISM score was 7.1±5.5. Eleven patients (92%) RACHS-1 score were ≥2. A large proportion of patients (10/12, 83.3%) had biventricular anatomy, among these patients atrioventricular septal defect was the most common diagnose. Two patients had single ventricle anatomy. The details of cardiac diagnosis and the cardiac surgery performed are summarized in Table 1. The median duration of mechanical ventilation was 60 days (range, 30-261 days) and the median length of PICU stay was 68 days (range, 30-301 days). The median duration of mechanical ventilation after tracheostomy was 16 days (range, 12-215 days). Demographics and patient characteristics are shown in Table 2.

Three patients who had a neurological impairment and 2 patients who had a failure of weaning underwent tracheostomy before extubation trials. Seven patients had at least 2 extubation trials. Diaphragmatic paralysis was the most common (5/12, 42%) indication for tracheostomy. Neurological impairment and hypotonicity were the next common indication and were present in 4 patients (33.3%). Direct laryngo-bronchoscopy was performed in 6 patients,

Table 1. Cardiac diagnoses, procedures and outcomes for patients requiring tracheostomy after congenital heart surgery and, the timing and indications for tracheostomy in all patients

Case no	Cardiac diagnosis	Primary surgery	Tracheostomy indication	Comorbidity	Number of extubation trials	Duration of intubation before tracheostomy (days)	Outcome
1	Balanced AVSD	AVSD repair	Diaphragm paralysis (bilateral)	-	3	14	Home ventilation
2	ASD, VSD, PDA	PDA closure, pulmonary banding	Diaphragm paralysis	DiGeorge syndrome, chronic pulmonary disease	-	50	Decannulated
3	DORV, PA, PFO, PDA, MBT shunt	Glenn	Prolonged PPV	-	2	18	Decannulated
4	AVSD	AVSD repair	Hypotonicity, diaphragm paralysis	Down syndrome	-	22	Home ventilation
5	ASD, VSD, PDA	PDA closure, pulmonary banding	Tracheobronchomalacia	Malnutrition	3	45	Decannulated
6	Single Ventricle, Glenn	Fontan procedure	Neurological impairment	-	-	6	Decannulated
7	ASD, VSD	AVSD repair, tricuspid valvuloplasty	Prolonged PPV	Down syndrome	3	67	Died
8	Tetralogy of Fallot	MBT shunt	Neurogical impairment	-	-	17	Trach collar
9	Shone's complex, MS	Mitral valvulopla sty	Bronchomalacia	-	4	46	Decannulated
10	ASD, VSD	AVSD repair	Diaphragm paralysis Neurological impairment Prolonged PPV	-	-	15	Home ventilation
11	Truncus arteriosus	Repair of truncus arteriosus	Tracheobronchomalacia	-	2	48	Trach collar
12	D-TGA	Senning procedure	Diaphragm paralysis chylothorax	-	3	28	Decannulated

AVSD: Atrioventricular septal defect, ASD: Atrial septal defect, VSD: Ventricular septal defect, PDA: Patent ductus arteriosus, DORV: Double outlet right ventricle, PA: Pulmonary atresia, PFO: Patent foramen ovale, MBT: Modified blalock-taussig, PPV: Positive pressure ventilation, MS: Mitral stenosis, D-TGA: Dextrotransposition of great arteries

Table 2. Demographics and patients characteristics of children requiring tracheostomy after operation for congenital heart disease

Characteristics	n=12
Male gender, n (%)	4 (33.3)
Median weight at surgery (kg, range)	6 (4-35)
Median operation age, day (IQR)	210 (75-262)
Chromosomal abnormalities, n (%)	3 (25)
PRISM score (mean, SD)	7.1 (5.5)
CPB time, minute (mean, SD)	73.2 (33.5)
Cross-clemp time, minute (mean, SD)	60.8 (34.3)
Time between surgery and tracheostomy, day, median (IQR)	25 (15-47)
Median duration of MV (day, range)	60 (30-261)
Duration of MV after tracheostomy (day, range)	16 (12-215)
Median length of PICU stay (day, range)	68 (30-301)
Median length of hospital stay (day)	81.5 (43-310)

IQR: Interquartile range, PRISM: The Pediatric Risk of Mortality, SD: Standard deviation, CPB: Cardiopulmonary bypass, MV: Mechanical ventilation, PICU: Pediatric intensive care unit

3 patients were diagnosed with tracheobronchomalacia (25%). In 2 patients, persistent cardiac insufficiency and prolonged mechanical ventilation were considered as indications for tracheostomy. Three patients (25%) had confirmed chromosomal anomalies which revealed trisomy 21 in two patients and DiGeorge syndrome in one patient. Two patients had chronic lung disease, one of them operated for tracheoesophageal fistula. Tracheostomy indications are summarized in Table 1. Genetic syndrome or at least one non-cardiac morbidity were present in 41.6% of patients.

No procedure-related complications have occurred in any of the patients during tracheostomy insertion. There were no mediastinal wound infections. Only one patient with DiGeorge syndrome had a tracheostomy site infection. Tracheitis and pneumonia were the most common complications. Before tracheostomy 10 patients (83.3%) had 14 VAP episodes whereas 7 patients (58.3%) had 9 VAP episodes after tracheostomy, although this difference was not significant (p=0.152). *Pseudomonas aeruginosa* was the most common pathogen (10/23, 43.4%) followed by *Klebsiella pneumoniae*

Table 3. Outcomes measures based on the timing of tracheostomy

	Tracheostomy day <30 day (n=7)	Tracheostomy day >30 day (n=5)	p-value
Duration of MV (day)	30 (28-60)	60 (60-261)	0.035
Duration of MV after tracheostomy (day)	22 (15-35)	12 (10-215)	0.456
Length of PICU stay (day)	40 (30-76)	69 (68-301)	0.059
Length of hospital stay (day)	51 (43-99)	84 (68-310)	0.089

MV: Mechanical ventilation, PICU: Pediatric intensive care unit

(6/23, 26%), *Stenotrophomonas maltophilia* (3/23, 13%) and *Acinetobacter baumannii* (2/23, 8.6%).

The number of patients who underwent tracheostomy placement <14 days and <30 days were 2 (16.6%) and 7 (58.3%) respectively. Comparing VAP episodes before and after tracheostomy in patients who underwent tracheostomy within 30 days of ventilation with those who underwent tracheostomy >30 days after intubation yielded no differences (p=1.000 and p=1.000, respectively). When evaluating the duration of mechanical ventilation patients in patients who had tracheostomy within 30 days there was a reduction in the median days of mechanical ventilation (30 vs. 60 days, p=0.035). Also between this group's duration of mechanical ventilation after tracheostomy, length of PICU stay and length of hospital stay yielded no differences (Table 3).

The median length of PICU stays after the tracheostomy was 41 days (range, 21-289 days). During the PICU stay, one patient died due to sepsis and multiorgan failure on the postoperative 47th day. Three patients were weaned from mechanical ventilation and successfully decannulated. Four patients (33%) were discharged home on mechanical ventilation and four patients (33%) on a trach collar. Two patients on a trach collar and one patient on mechanical ventilation were decannulated after PICU discharge. Among all patients with CHS undergoing tracheostomy, 6 (50%) of 12 were decannulated after a median time of 179 days (range, 34-463 days). The operative mortality was 8.3% (1/12) and the overall mortality during the first year of follow-up was 8.3% (1/12).

Discussion

In children with CHS, there is no consensus on the indications and optimal timing for tracheostomy. Although there is limited data on tracheostomy practices and outcomes in the pediatric population, recent studies reported that in PICU patients early tracheostomy may have significant benefits without adversly effecting mortality.[6] Early tracheostomy placement may shorten the length of PICU stay and reduce the incidence of VAP.[7] Our study population has a shorter time to tracheostomy and subjects who had tracheostomy within 30 days have a significantly shorter duration of mechanical ventilation. Two patients (patients 6 and 12) who underwent tracheostomy within 30 days, have been decannulated during their stay in PICU. The timing of tracheostomy varies according to the experience and approach of the clinician, and this may cause unnecessary or delayed tracheostomy procedures. In our cohort, there were no life-threatening complications related to tracheostomy.

Recent studies reported that patients with a history of cardiac surgery had a significantly longer duration of PICU admission to tracheostomy placement.[8,9] In a multicenter study that was evaluated to describe the use of tracheostomy, the median time between initiation of mechanical ventilation and tracheostomy placement was 14.4 days with significant variation in the primary diagnosis.[10] In literature, the median time for tracheostomy after CHS varies between 30-58 days.[11,12]

In this study, we identified a low rate of tracheostomy (0.5%) among patients after CHS and this result is comparable with previous studies. Although the incidence of tracheostomy after CHS is low, there is a significant increase over the years with possible attribution to the increased complexity of pediatric cardiac surgical procedures.[1,11] The incidence of tracheostomy after cardiac surgery has increased from 0.11% in 2000 to 0.76 in 2012, according to the STS congenital heart database.[1]

Infants and children undergoing cardiac surgery, especially patients with single ventricle physiology have a high risk for surgical complications and airway issues leading to prolonged mechanical ventilation. In a multicenter study that examined long-term mechanical ventilation and tracheostomy timing in PICU, they reported the majority of participants had underlying cardiac disease (57%) and 67% of those who underwent tracheostomy.[8] Published studies determined the several preoperative risk factors that are related to prolonged mechanical ventilation and the need for tracheostomy after CHS. Genetics and non-cardiac anomalies were present in 40-60% of the patients.[1,13] Additionally, postoperative morbidities including residual lesion, delayed sternal closure, cardiac arrest, sepsis and, airway issues related to the cardiovascular surgery may lead to prolonged mechanical ventilation and difficult weaning.[1,13,14] Hoskote et al.[12] described the postoperative

risk factors for tracheostomy after CHS as myocardial dysfunction (49%), tracheobronchomalacia (49%) and, diaphragmatic paralysis (35%). In our cohort, diaphragmatic paralysis was the most common indication for tracheostomy. Diaphragmatic plication is a successfull treatment option especially in infants with bilateral diaphragmatic paralysis.[15] Tracheobronchomalacia and prolonged mechanical ventilation due to neurological impairment were other indications for tracheostomy. Genetic syndrome or non-cardiac morbidities were present in 41.6% of patients. In 4 of 5 patients with diaphragm paralaysis, plication was not performed due to accompanying tracheostomy indication such as hypotonicity and neurological disorder that required positive pressure ventilatory support.

Although the rate of tracheostomy after CHS is increasing, tracheostomy requirement after CHS is still associated with a poor clinical course, high intra-hospital and extra-hospital mortality. In our cohort, the operative mortality was 8.3% that is lower than previously reported studies.[13,14] In our cohort, 11 patients (92%) RACHS-1 scores were ≥2 and 6 patients (50%) RACHS-1 scores were ≥3. Edwards et al.[16] reported children with more complex lesions and greater RACHS-1 scores had higher mortality rates. They reported the 5-year survival of 68% of children with home mechanical ventilation program after CHS, but the rate was only 12% in children with RACHS-1 of 4 or higher. However, in a study in which they analyzed the results of patients who needed tracheostomy and mechanical ventilation at home after CHS, they reported that there was no statistically significant difference in decannulation between patients with a RACHS-1 score >3 and patients with a RACHS-1 score ≤3.[17] In a large observational study, they demonstrated that subjects with congenital heart disease (CHD) had a 6.67 times higher risk of tracheostomy than those without CHD, and mortality risk was 3.8 times higher following tracheostomy in infants with CHD.[18]

In our study, the median length of PICU stay was 68 days. In our center, children requiring tracheostomy and mechanical ventilation are admitted in 5 beds intermediate unit facility. These patients have a high risk of death due to a tracheostomy-related complication after discharge. Therefore, in our clinic, the follow-up of patients with a high probability of decannulation and who may show reversibility for tracheostomy indication is followed up in this step-down unit. With this approach that patients can be followed for a longer period, it is aimed to prevent complications related to tracheostomy. In a single-center study, 5 out of 11 patients who underwent tracheostomy after CHS and died after initially being discharged home had tracheostomy-related complication.[11]

Study Limitations

There are some limitations of this study, as a result of it has retrospective design and a single-center study. As a result of the small number of patients in the study, a strong statistical evaluation could not be made. The study has a patient selection bias for age, the study cohort did not include the neonatal age group. Social conditions such as cooperation of the family, medical ward conditions may have affected the length of the PICU stay.

Conclusion

The early tracheostomy procedure facilitates the weaning process and shortens the duration of positive pressure ventilation. In this patient population, large scale studies are needed to identify risk factors for unsuccessful weaning and optimum timing for tracheostomy.

Ethics

Ethics Committee Approval: This study was approved by the institutional review boards with the permission for the use of patient data for publication purposes (21-1T/26, date: 07.01.2021 - Ege University Faculty of Medicine Medical Research Ethics Committee).

Informed Consent: Informed consent was received from the families.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Concept: P.Y.Ö., O.N.T., B.K., Design: P.Y.Ö., O.N.T., B.K., Data Collection or Processing: P.Y.Ö., E.E.T., İ.E., Analysis or Interpretation: P.Y.Ö., E.E.T., İ.E., Literature Search and Writing: P.Y.Ö.

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