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Survival Analysis of Surgical Tracheotomy in Pediatric Patients With Chronic Diseases in a Tertiary Health Center

Osman Erdoğan[1], Hatice Feray Arı[2], Betül Aktaş Kipoğlu[1], Sefa İncaz[1], Ahmet Yükkaldıran[3]

[1]Şanlıurfa Training and Research Hospital, Clinic of Otorhinolaryngology, Sanliurfa, Turkiye
[2]Adnan Menderes University Faculty of Medicine, Department of Pediatrics, Division of Pediatric Intensive Care, Aydin, Turkiye
[3]Private Lotus Hospital, Clinic of Otorhinolaryngology, Sanliurfa, Turkiye

ORCID ID: O.E. 0000-0001-9384-7881; H.F.A. 0000-0002-2208-2524; B.A.K. 0000-0002-2073-3784; S.İ. 0000-0002-1937-215X; A.Y. 0000-0002-1335-4110

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ABSTRACT

Objective: To investigate the effect of age group (infants and non-infants) and demographics on survival in pediatrics with tracheotomy.
Materials and Methods: Sex, age at the time of the tracheotomy, nationality, parental consanguinity, primary disease, date of discharge from the intensive care unit, and date of death were obtained from the medical records. The study population was categorized by nationality, parental consanguinity, and age group for survival analysis.
Results: The study included 140 pediatric patients who underwent a tracheotomy. The median age of the patients at the time of the tracheotomy was 1.23 years. The median follow-up after tracheotomy was 5.4 months. Consanguinity was present in 70.4% of the parents. Primary diseases were grouped as neurologic (37.1%), neuromuscular (29.3%), metabolic (12.1%), cardiopulmonary (8.6%), traumatic (6.4%), and syndromic (6.4%). Neuromuscular diseases were more common in infants, while neurologic and metabolic diseases were more common in non-infants. Eighteen patients were weaned and four patients were decannulated. The overall mortality rate was 70%. The median overall survival was 214 days. Infants (147 days) had a shorter survival than non-infants (286 days). Parental consanguinity and nationality did not affect survival.
Conclusion: In this study, among pediatric patients, infants are associated with a poor prognosis in survival. Neurologic and neuromuscular diseases may be thought to increase mortality among primary diseases. According to our study, it can be suggested that infants who cannot be decannulated due to neuromuscular diseases and are discharged with a tracheostomy are the group that should receive most attention regarding mortality among pediatrics with a tracheostomy.

Keywords: Infants, nationality, neurologic, parental consanguinity, pediatric tracheotomy, survival

INTRODUCTION

Tracheostomy is the surgical procedure used to open a window on the trachea. A tube is inserted into the window to create a new airway through which the patient is ventilated (1). This airway allows ventilation in the event of upper airway obstruction, more convenient removal of pulmonary secretions, and better management of ventilation in patients who required prolonged mechanical ventilation (2). An additional benefit is an increase in tissue oxygenation (3). Tracheotomy, previously indicated mainly for trauma and upper respiratory tract infections, is now often indicated for chronic diseases requiring prolonged mechanical ventilation (2).

Children with complex chronic diseases are more likely to be admitted to the intensive care unit (ICU) than children without those diseases. Length of stay and mortality in the ICU are higher in patients with complex chronic diseases (4). While the frequency of emergency tracheotomy has reduced in pediatrics, the rate of tracheotomy has increased due to better survival rates in the pediatrics and neonatal ICU (5). As the survival rate of children with congenital defects has improved, 41-63% of pediatric tracheotomies are performed on infants, and this rate has been increasing during the last few years (1,6).

This increasing number of tracheotomies prompted us to further investigate the demographics, chronic diseases, and survival of tracheostomized infants. The present study

Corresponding Author: Osman Erdogan **E-mail:** osman_erdogan@outlook.com
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compared the demographics, chronic diseases and, survival of pediatric patients, both infants and non-infants, at a tertiary healthcare center. The study is specific because the target population of our hospital is patients with low income.

MATERIAL AND METHODS

Study population

Ethics committee approval was obtained. All parents of the patients gave informed consent. The study comprised 140 pediatric patients who underwent tracheotomy in the pediatric ICU from August 2017 to December 2022. The inclusion criteria were age under 18 years at the time of tracheotomy and tracheotomy for prolonged intubation. Patients who underwent emergency tracheotomy for acute airway obstruction and patients with a history of previous tracheotomy were excluded from the study. The medical records of the patients were reviewed to determine gender, age at the time of tracheotomy, nationality, parental consanguinity, primary diseases, date of discharge from the ICU, and date of death. Data up to the last observation date, 31 December 2022, were used for survival analysis. All tracheotomies were performed by otolaryngologists at our hospital.

For statistical analysis, 63 patients up to 1 year of age were classified as infant group and 77 patients between 1 year and 18 years of age were classified as non-infant group. One hundred and eight patients were divided into two groups based on parental consanguineous status: consanguineous or nonconsanguineous. Patients were divided based on their nationality as either Syrian or Turkish.

Statistical analysis

Data analysis was performed using SPSS version 26.0 software (IBM Corp.; Armonk, NY, USA) statistical program. The distribution of the data was evaluated with the Shapiro-Wilk Test. For the analysis of non-parametric continuous variables, the Mann-Whitney U Test was used to compare independent groups. Pearson chi-squared test and Fisher exact test were used to compare categorical variables. The survival rate was calculated using the Kaplan-Meier method. The survival difference between the groups was evaluated by the log-rank test. The level of statistical significance was accepted as <0.05.

RESULTS

The median age of 140 patients at the time of tracheotomy was 1.23 years (interquartile range 0.50- 4.04 years, age range 1 month to 17.5 years). The analysis of the age distribution indicated a peak in infants, comprising of 45% of the patients (Figure 1). The male/female ratio was 1:1.26. The median follow-up period post-tracheotomy was 5.4 months (interquartile range 1.9 - 10.6 months) with the longest follow-up period being 59.5 months.

In the study, 109 patients were Turkish and 31 patients were Syrian. Of the patients, 76 had parental consanguinity, 32 did not, and for 32 of the patients, there was no information on consanguinity in the medical records. The most common chronic disease among patients was neurologic disease (37.1%), followed by neuromuscular disease (29.3%), metabolic disease (12.1%), cardiopulmonary disease (8.6%), trauma (6.4%), and syndromic disease (6.4%), as shown in Table 1.

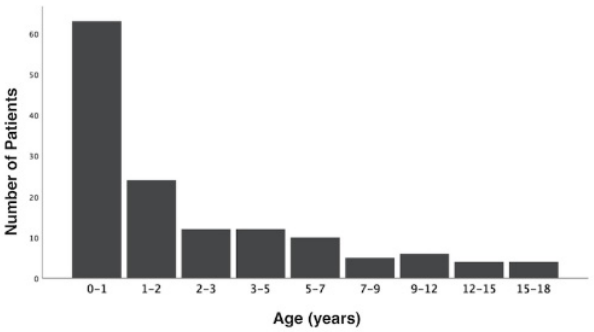

Figure 1: Age distributions of pediatrics at the time of tracheotomy

Table 1: Chronic diseases of the patients

Neurological disease (n, %)		**52 (37.1%)**
	Cerebral palsy	24
	Myelomeningocele	8
	Hypoxic ischemic encephalopathy	6
	Brain tumor	2
	West syndrome	2
	Hydrocephalus	2
	Herpes simplex virus encephalitis	1
	Encephalocele	1
	Metachromatic leukodystrophy	1
	Krabbe disease	1
	Holoprosencephaly	1
	Leigh syndrome	1
	Corpus callosum agenesis	1
	Canavan disease	1
Neuromuscular disease (n, %)		**41 (29.3%)**
	Spinal muscular atrophy	31
	Hypotonic infant	5
	Muscular dystrophy	3
	Mitochondrial myopathy	1
	Myasthenia gravis	1
Metabolic disease (n, %)		**17 (12.1%)**
	Neurometabolic Disease	11
	Mucopolysaccharidosis	1
	Congenital lactic acidosis	1
	Glutaric aciduria type 1	1

Table 1: Continue

Metabolic disease (n, %)		**17 (12.1%)**
	Tay-Sachs disease	1
	I-cell disease	1
	Sandhoff disease	1
Cardiopulmonary disease (n, %)		**12 (8.6%)**
	Asphyxia	7
	Bronchiolitis	2
	Laryngomalacia	2
	Foreign body aspiration	1
Syndromic disease (n, %)		**9 (6.4%)**
	Down syndrome	3
	Pierre Robin sequence	2
	DiGeorge syndrome	1
	Miscellaneous	3
Trauma (n, %)		**9 (6.4%)**
	Head trauma	5
	Traffic accident	4
Total (n, %)		**140 (100%)**

During the follow-up period, 18 patients were successfully weaned from mechanical ventilation. The median duration for successful weaning was 21 days (12-30.5 IQR). The median time to decannulation was 54 days (20.25-84 IQR) in four patients. None of the patients with neurologic and neuromuscular diseases were able to be decannulated. Fifty patients (35.7%) were discharged from the hospital. The median duration from tracheostomy to discharge was 73 days (30.25-132 IQR).

The overall mortality rate during follow-up was 70%, with 78% observed in infants and 64% in non-infants. No deaths were attributed to tracheotomy. When comparing patients who died to those who were alive during the last observation, there was no difference between the groups regarding their sex, nationality, chronic diseases, and tracheotomy timing (Table 2).

The analysis of infants and non-infants showed a tendency for more females in the infant group and more males in the non-infant group (p=0.037). No difference was found between the groups in terms of nationality, parental consanguinity, discharge rates, discharge times, and tracheotomy timing. The primary diagnosis showed a statistically significant difference between groups (Table 3). Post-hoc tests indicated a greater occurrence of neuromuscular disease in the infant group and a greater occurrence of neurological and metabolic disease in the non-infant group (Figure 2).

The survival analysis revealed a median survival of 214 (166-262 95% CI) days for all patients. The 1-year and 3-year survival rates for these patients were 0.319 and 0.158, respectively (Figure 3).

The median survival time for infant and non-infant groups was 147 days (119-175 95% CI) and 286 days (190-382 95% CI), respectively (Figure 4). Non-infants had a higher overall survival rate than infants (p=0.005). The survival rates at one year were 0.192 for the infants and 0.415 for the non-infants. Survival rates at three years were 0.076 and 0.217 in infants and non-infants, respectively.

Survival analysis based on parental consanguinity showed that the median survival was 214 (158-270 95% CI) days in patients with consanguineous parents and 285 (0-652 95% CI) days in patients without consanguineous parents (Figure 5). There was no statistically significant difference between these groups (p=0.084).

Table 2: Chronic diseases and demographics of the pediatric patients who were alive at last observation and died

	Alive at last observation (n=42)	Died (n=98)	p
Sex (n, %)			**0.373**
Female	21 (33.9)	41 (66.1)	
Male	21 (26.9)	57 (73.1)	
Nationality (n, %)			**0.564**
Turkish	34 (31.2)	75 (68.8)	
Syrian	8 (25.8)	23 (74.2)	
Primary diagnosis (n, %)			**0.108**
Neurological disease	17 (32.7)	35 (67.3)	
Neuromuscular disease	8 (19.5)	33 (80.5)	
Neurometabolic disease	8 (47.1)	9 (52.9)	
Cardiopulmonary disease	3 (25)	9 (75)	
Syndromic disease	1 (11.1)	8 (88.9)	
Trauma	5 (55.6)	4 (44.4)	
Tracheotomy timing, day (median, IQR)	319 (152 - 672)	146 (40 - 261)	0.752

IQR: interquartile range

Table 3: Chronic diseases and demographics of the infants and non-infants

	Infants (n=63)	Non-infants (n=77)	p
Sex (n, %)			**0.037**
Female	34 (54%)	28 (36%)	
Male	29 (46%)	49 (64%)	
Nationality (n, %)			**0.402**
Turkish	47 (75%)	62 (81%)	
Syrian	16 (25%)	15 (19%)	
Parental consanguinity (n, %)			**0.711**
Yes	28 (44%)	48 (62%)	
No	13 (21%)	19 (25%)	
Unknown	22 (35%)	10 (13%)	
Primary diagnosis (n, %)			**<0.001**
Neurologic disease	15 (23.8%)	37 (48.1%)	
Neuromuscular disease	30 (47.6%)	11 (14.3%)	
Trauma	2 (3.2%)	7 (9.1%)	
Metabolic disease	5 (7.9%)	12 (15.6%)	
Syndromic disease	7 (11.1%)	2 (2.6%)	
Cardiopulmonary disease	4 (6.3%)	8 (10.4%)	
Tracheotomy timing, day (median, IQR)	39 (27.0-76.0)	38 (24.5-73.5)	0.744
Discharged patients	22 (34.9%)	28 (36.4%)	0.859
Discharge timing, day (median, IQR)	73 (34-132)	65 (28.75-48.25)	0.953

IQR: interquartile range

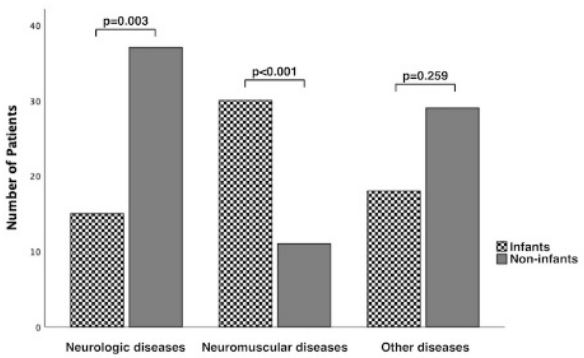

Figure 2: The bar graph represents the relationship between age groups (infants and non-infants) and primary diseases. Note that the p values obtained from post-hoc tests after the chi-square test

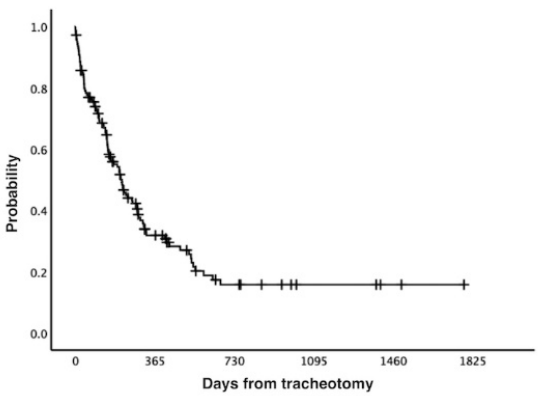

Figure 3: Kaplan-Meier survival curve of all patients after the time of tracheotomy

Survival analysis based on nationality showed that the median survival was 215 (172-258 95% CI) days in Turkish patients and 163 (69-257 95% CI) days in Syrian patients. Statistically significant differences in survival were not found between nationalities (p=0.693).

DISCUSSION

In this study, infants with tracheotomy had a lower survival than non-infants with tracheotomy. Non-infants were more likely to have neurologic and metabolic diseases, whereas infants were more likely to have neuromuscular diseases, thus primary diseases may be one of the factors that could cause the difference in survival.

Our study population had a high prevalence of neurological and neuromuscular diseases. Consanguineous marriage and low income status are risk factors for neurologic diseases (e.g., SMA and cerebral palsy) (7–9). Our hospital has patients from low-income populations and is located in the province with

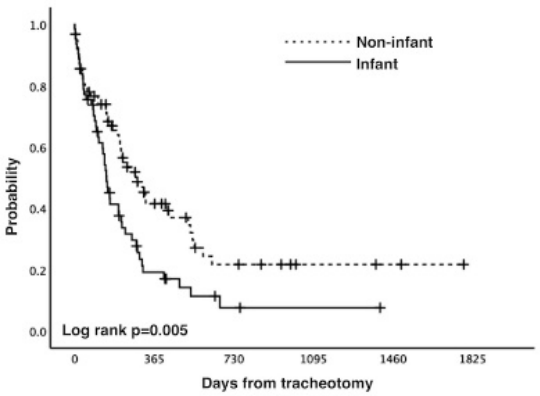

Figure 4: Kaplan-Meier survival curve of infants and non-infants after the time of tracheotomy

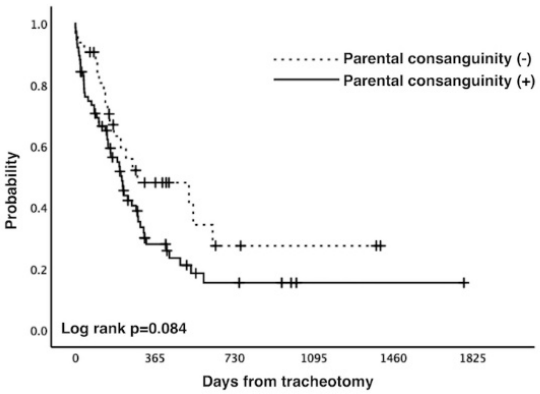

Figure 5: Kaplan-Meier survival curve of parental consanguinity status after the time of tracheotomy

the highest rate of consanguineous marriages (18.4%) in the country. A significantly elevated incidence of neurologic and neuromuscular conditions in our study was probably due to a very high rate of parental consanguinity (70%). Patients with parental consanguinity demonstrated a lower survival rate, although not statistically significant (p=0.084).

The indications for tracheotomy have changed in recent years from air-way compromises to neurologic diseases requiring prolonged ventilation (10). Additionally, patients who underwent tracheostomy due to neurological reasons have lower decannulation rates and a higher risk of mortality after being discharged (11). Therefore, analyzing the survival of tracheotomized pediatrics after discharge from the ICU and determining the prevalence of neurologic disease may provide a different perspective on the interpretation of mortality reported in the literature. Hebbar et al. revealed that the occurrence of neurological diseases was 24%, and the mortality rate was 27% (12). A 10-year study by McPherson et al. of pediatric tracheostomies found neurologic conditions were associated with higher mortality and lower decannulation rates than airway conditions (13). The study reported a neurological disease rate of 52% and a mortality rate of 23%. According to Funamura et al., bronchopulmonary dysplasia and congenital heart diseases were found to be predictors of mortality (14). Despite the rate of neurologic disease being 31.6%, their overall mortality rate (16.6%) was lower in comparison to the above studies. Salley et al. reported 16% mortality in patients under 3 years of age in their survival analysis, most of whom had respiratory failure (15). Akangire et al. conducted a study that found an overall mortality rate of 21%. The study also revealed that 70.6% of the reasons for tracheotomies were respiratory, and 6.86% were neurological (16). According to Cristea et al., the study revealed an 18.6% mortality rate in patients with severe bronchopulmonary dysplasia (17).

In our study, the mortality rate was 70%. The most common chronic disease group was neurological/neuromuscular disease (66%). In this context, it seems reasonable to assume that the mortality rate may increase as the primary diseases of patients shift from respiratory diseases to neurologic and neuromuscular diseases. Possible reasons for the high mortality found in this study are: (1) A high number of pediatric patients with complicated diseases were referred to our hospital from nearby hospitals, (2) Neurologic and neuromuscular diseases are prevalent in our ICU, (3) The low income status affecting home care, and (4) High rate of consanguineous marriages.

Evaluating the Kaplan-Meier overall survival curve in our study, most deaths occurred between 6-12 months after tracheotomy (Figure 2). Similar to our study, Hebbar et al. reported that although most of the primary diseases were cardiopulmonary diseases, the greatest decrease in survival occurred between 6-12 months in the survival analysis (12). They proposed that progression of the underlying disease, comorbidities, cannula dislocation, cannula plugging, tracheotomy-related accidents (e.g., bleeding), and inadequate post-discharge care are the factors leading to this situation. Despite the low discharge rate in our study, progressive neurologic, neuromuscular, and metabolic disease and increased pulmonary problems due to the low decannulation rate may have resulted in the greatest decrease in survival between 6 and 12 months.

Neonatal survival has increased over the years owing to the improvement in ICUs. These improvements led to an increase in congenital anomalies, followed by a rise in the rate of tracheotomy in infants from 41% to 63% in pediatrics (1,6). Therefore, infants deserve special consideration regarding tracheotomy outcomes in pediatrics. Berry et al. found higher in-hospital mortality in infants compared to patients aged 1-4 years in each year from 1997 to 2006 in their retrospective study of 18806 tracheotomized patients (18). Similarly, Sakai et al. reported increased overall mortality in the infant group in pediatrics (19). Our study also reveals that infants exhibited lower survival rates than non-infants.

This study has some limitations. In infants, mortality rates are higher when there is low birth weight and preterm birth (18,20). Since our hospital received a large number of referred patients from other hospitals and refugee patients from cross-border, factors such as preterm birth and low birth weight could not be analyzed in the study because the birth information of

most patients could not be reached. Furthermore, concerning the timing of tracheotomy, the delay in parental informal consent may have influenced the survival outcome of this study. The high mortality rate of our research, as opposed to the literature, demonstrates the strength of our study. Hence, this has highlighted that different socio-demographic characteristics may lead to different survival outcomes. Based on the limitations of this study, it is recommended that future studies on the survival of pediatrics with tracheotomy should examine sociodemographic characteristics in a thorough and comprehensive way.

CONCLUSION

The result of the present study showing that tracheotomized infants have a lower survival compared to tracheotomized non-infants is valuable because infants have become a peak group of pediatrics requiring tracheotomy in recent years. Nowadays, tracheotomy-related mortality has decreased in infants, whereas the risk of mortality after discharge increases due to the primary disease or the progression of primary disease, especially in neurologic and neuromuscular diseases. Therefore, family education on home care is critical for the caregivers of infants. In addition, if needed due to sociodemographic characteristics, the organization of educational programs on issues such as consanguineous marriage and prenatal diagnosis is important to reduce the frequency of diseases that predispose to tracheotomy in infants.

Ethics Committee Approval: This study was approved by Harran University Clinical Research Ethics Committee (Date: 21.03.2022, No: 06).

Informed Consent: Written informed consent was obtained.

Peer Review: Externally peer-reviewed.

Author Contributions: Conception/Design of Study- O.E., H.F.A., B.A.K., S.İ., A.Y.; Data Acquisition- O.E., H.F.A., B.A.K., S.İ., A.Y.; Data Analysis/Interpretation- O.E., H.F.A.; Drafting Manuscript- O.E., H.F.A.; Critical Revision of Manuscript- O.E., H.F.A., B.A.K., S.İ., A.Y.; Final Approval and Accountability- O.E., H.F.A., B.A.K., S.İ., A.Y.; Material or Technical Support- O.E., H.F.A.; Supervision- O.E., H.F.A., B.A.K., S.İ., A.Y.

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