AMERICAN ACADEMY OF OTOLARYNGOLOGY–HEAD AND NECK SURGERY FOUNDATION

Original Research—Pediatric Otolaryngology

Otolaryngology–Head and Neck Surgery
2025, Vol. 00(00) 1–10

DOI: 10.1002/ohn.1248
http://otojournal.org
WILEY

Tracheostomy Outcomes in Children With Bronchopulmonary Dysplasia

Ada Cleary Sher, BS[1,2], Humra Shamim, MS[1], Jemma Maynard, BA[3], Jacob Stack, BS[1], Isaac Kistler, MS[2,3,4], Megan McNutt, BS[3], Hajera Afreen, BA[2,3], Amy Manning, MD[3], Audrey Miller, MD[5], Prasanth Pattisapu, MD, MPH[3], and Tendy Chiang, MD[1,3]

Abstract

Objective. To quantify the tracheostomy-related morbidity and mortality, readmissions, and airway interventions in tracheostomy-dependent children with bronchopulmonary dysplasia (BPD).

Study Design. Retrospective chart review.

Setting. Tertiary care children's hospital.

Methods. Infants with BPD who received tracheostomy by an otolaryngologist between January 2016 and December 2022 at a single institution were included. Surviving patients were followed to at least 2 years of age. Data were extracted from electronic medical records for patient characteristics, clinical encounters, and surgical visits.

Results. There were 76 patients included in this study. The overall mortality was 30.3% (23/76) with one tracheostomy-related death. Tracheostomy occurred at a median 56 weeks postmenstrual age (PMA). Tracheitis was the most common short- and long-term adverse event (within the first postoperative week, 21%; after postoperative day 7, 81%). Other complications observed include stomal granuloma (77%), airway stenosis (69%), and accidental decannulation (38%). The 30-day and 2-year readmission rates were 32% and 61%, of which 61% and 76% were for tracheostomy-related causes, respectively. The most common reason for readmission over these time periods was tracheitis. Overall decannulation rate for surviving patients was 68%, and decannulation occurred at a median 3.1 years of age. Most surviving patients returned for operative airway intervention (85%); 43% required open airway reconstruction.

Conclusion. Tracheostomy-related morbidity was low in this cohort despite a majority of patients experiencing multiple tracheostomy-related adverse events and readmissions. Patient comorbid conditions, BPD severity, and socioeconomic status were not significantly associated with outcomes. Larger studies are needed to assess the airway interventions and outcomes in this population.

Keywords

BPD, bronchopulmonary dysplasia, ENT, tracheostomy

Received August 12, 2024; accepted March 20, 2025.

Bronchopulmonary dysplasia (BPD) is the most common morbidity of prematurity, with an annual incidence of 10,000 to 15,000 infants per year in the United States.[1] BPD affects both the small and large airways and is characterized by heterogeneous lungs with alveolar simplification and abnormal pulmonary vasculature.[2,3] With the improved overall survival of infants with BPD due to advancements in clinical management, including surfactant and postnatal steroid administration, the rate of chronically ventilated patients has increased. As a result, tracheostomy rates have also increased, and tracheostomy is performed in approximately 23% of all infants with BPD, although this rate varies depending on the center.[4]

Although the population of tracheostomy-dependent infants with BPD is increasing, data on tracheostomy outcomes, specifically surgical interventions, are limited. Specifically, the readmission rates, tracheostomy-related complications, as well as the prevalence of laryngotracheal stenosis, tracheobronchomalacia, and subsequent airway surgery are not well understood. Thus, the

[1]Center for Regenerative Medicine, Abigail Wexner Research Institute, Nationwide Children's Hospital, Columbus, Ohio, USA
[2]The Ohio State University College of Medicine, Columbus, Ohio, USA
[3]Department of Pediatric Otolaryngology, Nationwide Children's Hospital, Columbus, Ohio, USA
[4]Biostatistics Resource at Nationwide Children's Hospital, Columbus, Ohio, USA
[5]Department of Pediatrics, Division of Neonatology, Nationwide Children's Hospital, Columbus, Ohio, USA

Corresponding Author:
Tendy Chiang, Center for Regenerative Medicine, Abigail Wexner Research Institute, Nationwide Children's Hospital, Columbus, OH, USA.
Email: Tendy.Chiang@nationwidechildrens.org

primary objective of this study was to determine the tracheostomy-related outcomes, readmissions, and airway interventions in tracheostomy-dependent children with BPD. In terms of outcomes, we hypothesize that patient survival and rehospitalizations would be negatively associated with increasing medical complexity and lower socioeconomic status. Understanding these outcomes can help guide counseling and long-term airway surveillance for this population.

Materials and Methods

This retrospective study was approved by the Nationwide Children's Hospital Institutional Review Board before the start of data collection. We performed a retrospective single-center cohort study of children with BPD who underwent tracheostomy between January 1, 2016, and December 31, 2022. The institution's electronic medical record system was queried for patients with BPD and tracheostomy (defined by International Classification of Disease 10 and Current Procedural Terminology/ Healthcare Common Procedure Coding System codes). All pediatric patients diagnosed with BPD who received tracheostomy by a pediatric otolaryngologist at a single tertiary care children's hospital were included. BPD diagnosis was verified using Jensen criteria and was applied for patients born <32 weeks gestational age (GA).[5] Surviving patients were followed until at least 2 years of age. Patients who did not meet BPD diagnosis criteria, had insufficient follow-up, and/or did not have a tracheostomy performed at our institution were excluded.

Electronic medical records were reviewed for demographic information, comorbid conditions, age at tracheostomy, age at initial hospital discharge, postoperative adverse events, unplanned hospital readmissions, all operative airway surveillance and/or interventions, and age at decannulation. To assess the socioeconomic status of patients, patient zip codes were obtained, and the childhood opportunity index (COI) scores were generated using data from 2019. Tracheitis in this study was defined by increased tracheal secretions with a diagnosis of tracheitis by a clinician and subsequent antibiotic treatment. Tracheostomy-related mortality was defined by death caused by accidental decannulation, mucous plugging or obstruction, sepsis from tracheal infection, false tract, tracheoesophageal fistula, or tracheoinnominate fistula. Surgical airway interventions were recorded for all patients. Airway findings in surviving patients were obtained from operative notes after tracheostomy until the most recent follow-up or airway reconstruction.

Statistical Analysis

Demographic characteristics were summarized using count and percentage for categorical variables and median and interquartile range (IQR; 25th and 75th percentile) for continuous variables. Univariable logistic regression models were used to estimate odds ratios for outcomes (survival or 30-day readmission) with exposure variables (patient characteristics, adverse events, and age at discharge). Estimates and 95% confidence intervals are presented with *P*-values; no adjustments were made to control for multiple comparisons. All analyses were done in R version 4.3.

Results

Our center cares for a large volume of patients with BPD, and 76 patients met the inclusion criteria as defined by the diagnosis of BPD and presence of an existing tracheostomy (**Figure 1**). Patient characteristics and demographics are summarized in **Table 1**. In terms of patient socioeconomic status, 62% of patients (n = 46/76) were from zip codes with low or very low COI scores (**Table 2**). Specific rates of comorbid conditions are given in Supplemental Table S1, available online. The median

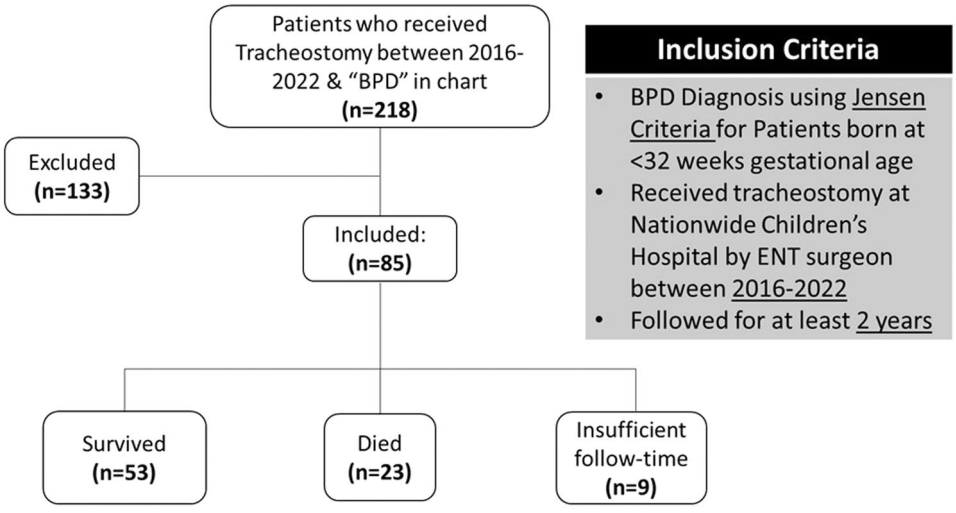

Figure 1. Flowchart depicting the inclusion of 76 patients born at <32 weeks gestational age, diagnosed with BPD who received tracheostomy at a single institution. Nine patients were lost to follow-up.

age at tracheostomy was 56 weeks postmenstrual age (PMA) (IQR 50-62). Ninety-three percent (93%, n = 71/76) of patients received tracheostomy before primary discharge. Fifty-six patients were discharged (74%) at a median age of 80.9 weeks PMA (IQR 63-102). The median time from tracheostomy to discharge was 34 weeks (IQR 17-43). Out of patients discharged, 5.4% (n = 3) of patients were discharged to foster care, and 7.1% (n = 4) were discharged to long-term care facilities. At primary discharge, 49% (n = 25) of patients were discharged with home mechanical ventilation, 47% (n = 24) were discharged on supplemental oxygen, and 4% (n = 2) were discharged on room air.

Table 1. Patient Characteristics

Characteristics	No. or median	IQR or %
Sex, male	44	58
Race		
White	40	53
Black or African American	25	33
Asian	1	1.3
Multiracial	9	12
Ethnicity, Hispanic	1	1.3
Gestational age at birth, wk	26.64	25.00, 28.14
Birth weight, kg	0.78	0.59, 1.00
Jensen grade (BPD severity)		
1 (mild)	5	11
2	13	28
3 (severe)	29	62
Comorbid conditions[a]		
1-2	13	19
3-4	38	57
>4	25	37

Abbreviations: BPD, bronchopulmonary dysplasia; IQR, interquartile range; IUGR, intrauterine growth retardation; NEC, necrotizing enterocolitis; SGA, small for gestational age.
[a]Comorbid conditions: IUGR, SGA, NEC, pulmonary hypertension, cardiac condition, neurologic condition, metabolic condition, and genetic syndromes.

Tracheostomy-Related Mortality Was Rare Despite High Overall Mortality

The median length of follow-up for all patients was 3.9 years (IQR 2.3-6.1). The overall mortality rate was 30.3% (n = 23); 78% of these patients (n = 18/23) died during primary admission, most commonly from severe BPD with subsequent withdrawal of care (Supplemental Table S2, available online). The median age at death was 1.6 years PMA (IQR 1.4-2). There were no tracheostomy-related deaths during primary hospitalization. One (1.7%) tracheostomy-related death was identified in the entire cohort, resulting from accidental decannulation at home. When comparing the number of comorbid conditions, BPD severity, and COI of patients who survived versus those who died, no significant differences were observed (**Table 2**).

Tracheitis Was the Most Common Tracheostomy-Related Adverse Event and Was a Common Reason for Readmission

In the early postoperative period (ie, the first week following tracheostomy), 50% of patients (38/76) had at least one tracheostomy-related adverse event and in the late postoperative period (beyond the first week to the end of surveillance period), 94% of surviving patients (n = 50/53) had at least one adverse event with patients experiencing a median of four types of adverse events after the first postoperative week (**Table 3**). The most common adverse event was tracheitis with early and late rates of 21% (n = 16) and 81% (n = 43), respectively (**Table 3**). Following discharge, the 30-day unplanned readmission rate was 32% (n = 18/56). Of the 18 patients

Table 2. Patient Characteristics by Outcome

Characteristic	Overall (n = 76)[a]	Expired (n = 23)[a]	Survived (n = 53)[a]	OR[b]	95% CI[b]	*P*-value
Comorbid conditions[c]	4.00 (3.00, 5.00)	4.00 (3.00, 5.50)	4.00 (3.00, 5.00)	1.31	0.95, 1.85	.11
Severe BPD[d]	53 (71%)	19 (86%)	34 (64%)	3.54	1.03, 16.4	.065
COI[e]						
Very high	9 (12%)	3 (13%)	6 (12%)	-	-	
High	10 (14%)	2 (8.7%)	8 (16%)	0.50	0.05, 3.96	.5
Moderate	9 (12%)	3 (13%)	6 (12%)	1.00	0.13, 7.50	>.9
Low	24 (32%)	10 (43%)	14 (27%)	1.43	0.30, 8.07	.7
Very low	22 (30%)	5 (22%)	17 (33%)	0.59	0.11, 3.56	.5

Abbreviations: BPD, bronchopulmonary dysplasia; IQR, interquartile range; IUGR, intrauterine growth retardation; NEC, necrotizing enterocolitis; SGA, small for gestational age.
[a]Median (IQR); n (%).
[b]OR = odds ratio, CI = confidence interval.
[c]Comorbid conditions: IUGR, SGA, NEC, pulmonary hypertension, cardiac condition, neurologic condition, metabolic condition, and genetic syndromes.
[d]n = 75.
[e]Childhood opportunity index; n = 74.

Table 3. Tracheostomy-Related Adverse Events of Patients[a]

Adverse event	Early (n = 76)[b]	Late (n = 53)[b]
Tracheitis	16 (21)	43 (81)
Stomal wound	13 (17)	6 (11)
Pneumothorax	5 (7)	4 (8)
Bleeding from stoma	5 (7)	16 (30)
Accidental decannulation	4 (5)	20 (38)
Mucous plug	4 (5)	-
Pneumonia	4 (5)	-
Death	0 (0)	1 (2)
Stomal granuloma	-	41 (77)
Airway stenosis[c]	-	36 (69)
Grade I SGS	-	9 (17)
Grade II SGS	-	5 (10)
Grade III SGS	-	11 (21)
Grade IV SGS	-	1 (2)
Tracheal stenosis	-	20 (38)
TCF requiring surgical closure	-	17 (32)
TEF	-	3 (6)
TIF	-	0 (0)

Abbreviations: SGS, subglottic stenosis; TCF, tracheocutaneous fistula; TEF, tracheoesophageal fistula; TIF, tracheoinnominate fistula.
[a]Early events occurred in the first postoperative week. Late events occurred any time after postoperative day 7. A "-" indicates event not measured.
[b]n (%).
[c]n = 52.

Table 4. Unplanned Readmissions[a]

Readmission causes	30 days (n = 21)[b]	30 days to 2 years (n = 121)[b]
*Tracheitis	10 (48)	43 (36)
Respiratory distress (viral URI and/or BPD exacerbation)	5 (24)	26 (21)
Lack of caregiver	2 (10)	3 (2)
*Accidental decannulation	1 (5)	3 (2)
*Bloody secretions from trach	1 (5)	4 (3)
G-tube displacement	1 (5)	0 (0)
Ventilator setting adjustment	1 (5)	0 (0)
Gastrointestinal	0 (0)	16 (13)
Pneumonia	0 (0)	3 (2)
Hypothermia	0 (0)	3 (2)
*Mucous plug	0 (0)	1 (1)
Benzodiazepine withdrawal	0 (0)	1 (1)
Pulmonary hypertension	0 (0)	1 (1)
Sepsis workup	0 (0)	1 (1)
*Blood clot in trach	0 (0)	1 (1)
Hydrocephalus	0 (0)	1 (1)
Chest wall abscess	0 (0)	1 (1)
Femur fracture	0 (0)	1 (1)
Parental concerns	0 (0)	1 (1)
*Increased tracheal secretions	0 (0)	1 (1)
Desaturations	0 (0)	1 (1)
*Stomal hemorrhage	0 (0)	1 (1)
Failure to thrive	0 (0)	1 (1)

Abbreviation: BPD, bronchopulmonary dysplasia.
[a]Eighteen patients were readmitted 21 times within 30 days after primary discharge. Thirty-three patients were readmitted 121 times between 30 days and 2 years after primary discharge. Tracheostomy-related causes are noted with a *.
[b]n (%).

readmitted over 30 days, 61% (n = 11) of patients were readmitted for tracheostomy-related causes. The most common reasons for early readmission were tracheitis (n = 10, 48%), BPD exacerbation (n = 5, 19%), and lack of caregiver (n = 2, 10%, **Table 4**). In the first 2 years following primary discharge, 61% of living patients (33/54) were readmitted with a median of two unplanned readmissions/2 years (IQR 1-6); 76% (n = 41/54) of these patients had at least one readmission for a tracheostomy-related cause, the most common of which was tracheitis (n = 43, 36%, **Table 4**). Patients who were readmitted in the first 30 days following discharge had significantly fewer comorbid conditions (n = 11/56, 3 comorbid conditions vs n = 45/56, 4 comorbid conditions $P = .034$, **Table 5**). There were no significant differences in readmissions when comparing COI, age at discharge, or BPD severity in this cohort (**Table 5**).

Posttracheostomy Airway Stenosis and Malacia Were Commonly Observed With Open Airway Reconstruction Required in 43% (n = 23/53) of the Surviving Population

Airway findings and interventions were documented in surviving patients. Overall, 98% of patients (n = 52/53) returned to the operating room (OR) for airway surveillance and/or intervention following tracheostomy placement for a median of 1.1 times per year (IQR 0.5-1.9). During bronchoscopy surveillance, 3.8% (n = 2/52) had a subglottic cyst, and one patient (1.9%) had a posterior tracheal wall diverticulum. Notably, 69% (n = 36/52) of patients had a type of laryngotracheal stenosis. Specifically, 52% (n = 27/52) had subglottic stenosis (SGS) with 23% (n = 12/52) of them being grade 3 or 4. Tracheal stenosis was observed in 38% of patients (n = 20/52) with A-frame deformities as the most common finding. Suprastomal collapse was common and seen in 58% of this group (n = 30/52). Tracheomalacia was seen in 35% of patients (n = 18/52) and bronchomalacia was seen in 33% (n = 17/52) with 23% having tracheobronchomalacia (n = 12/52). Of the patients who returned to the OR, 87% received some form of airway intervention (45/52, **Table 6**). Overall, 43% of patients (n = 23/53) required open airway reconstruction.

68% of Surviving Patients Were Decannulated at a Median 3.1 Years of Age

Overall, 68% of surviving patients (n = 36/53) were decannulated at a median age of 3.1 years (IQR

Table 5. Patient Characteristics by 30-Day Readmission Status

Characteristic	Overall (n = 56)[a]	Not readmitted (n = 45)[a]	Readmitted (n = 11)[a]	n	OR[b]	95% CI[b]	*P*-value
Comorbid conditions[c]	4.00 (3.00, 5.00)	4.00 (3.00, 5.00)	3.00 (1.50, 4.50)	56	0.56	0.31, 0.92	.034*
PMA at discharge	81 (63, 101)	87 (65, 108)	66 (62, 78)	56	0.97	0.93, 1.00	.051
Severe BPD	36 (65%)	28 (64%)	8 (73%)	55	1.52	0.38, 7.71	.6
COI[d]				54			
Very high	7 (13%)	6 (14%)	1 (9.1%)		-	-	
High	8 (15%)	7 (16%)	1 (9.1%)		0.86	0.03, 25.0	>.9
Moderate	6 (11%)	5 (12%)	1 (9.1%)		1.20	0.04, 36.0	>.9
Low	16 (30%)	14 (33%)	2 (18%)		0.86	0.07, 20.5	>.9
Very low	17 (31%)	11 (26%)	6 (55%)		3.27	0.41, 69.5	.3
Early adverse event				56	0.91	0.33, 2.12	.8
0	31 (55%)	25 (56%)	6 (55%)				
1	18 (32%)	14 (31%)	4 (36%)				
2	6 (11%)	5 (11%)	1 (9.1%)				
3	1 (1.8%)	1 (2.2%)	0 (0%)				
Late adverse event	4.00 (3.00, 5.00)	4.00 (3.00, 5.00)	4.50 (3.00, 6.00)	51	1.19	0.81, 1.84	.4
ENT OR trips	6.0 (2.0, 9.3)	5.0 (2.0, 9.0)	8.0 (5.0, 9.5)	56	1.08	0.94, 1.24	.3

Abbreviations: BPD, bronchopulmonary dysplasia; ENT OR, ear, nose, and throat operating room; IUGR, intrauterine growth retardation; NEC, necrotizing enterocolitis; PMA, postmenstrual age; SGA, small for gestational age.
*$P < .05$.
[a]Median (IQR); n (%).
[b]OR = odds ratio, CI = confidence interval.
[c]Comorbid conditions: IUGR, SGA, NEC, pulmonary hypertension, cardiac condition, neurologic condition, metabolic condition, and genetic syndromes.
[d]Childhood opportunity index.

Table 6. Ear, Nose, and Throat Interventions Performed on Surviving Patients (n = 53)

Intervention	n (%)
Granuloma removal	36 (68)
Dilation	19 (36)
Laryngotracheal reconstruction (LTR)	17 (32)
Single-stage LTR	7 (13)
Kenalog injection	10 (19)
Slide tracheoplasty	4 (8)
Tracheal resection	2 (4)

2.5-4.1), and a median of 2.6 years (IQR 2.1-3.6) after tracheostomy. The median age of the 17 patients (32%) not decannulated at follow-up was 4.0 years (IQR 2.6-6.3). When comparing characteristics between patients who were decannulated at 3 years versus those who were not, no significant differences were observed (**Table 7**). The reasons for continued tracheostomy-dependence at 3 years of age are summarized in Supplemental Table S3, available online. A summary of tracheostomy outcomes for each patient is given in **Figure 2**.

Discussion

Tracheostomy-dependent children with BPD represent a medically complex patient population, as highlighted in our cohort. The median number of additional comorbid conditions in this group was high with 71% (n = 54) of patients having four or more additional comorbid conditions. We speculate that these comorbid conditions, including cardiac, neurologic, and genetic conditions, could impact these listed outcomes, including mortality and time to decannulation. A majority of patients were diagnosed with grade III (severe) BPD by Jensen criteria, meaning they required mechanical ventilation at 36 weeks PMA. This is consistent with existing literature, with one study reporting that patients with grade III BPD were 6 times as likely to receive tracheostomy.[4] Our patient population meets many priorly reported risk factors for receiving tracheostomy in BPD patients, including male sex, extreme prematurity (less than 28 weeks GA), weight less than 1 kg at birth, and intrauterine growth restriction.[6,7]

Our cohort also demonstrated sociodemographic complexity. A majority of infants were white with an overrepresentation of black infants. An increased rate of tracheostomy in black infants with BPD has been reported in the literature with one study finding that black infants were 25% more likely to receive tracheostomy compared to white infants.[8] A majority of patients in this study also were from areas with low or very low COI. The overlap of poor socioeconomic status and high patient complexity in this group can be seen in the relatively high rate of readmissions for lack of caregiver availability, with 9% of surviving patients being readmitted for this reason. Parents and family members of tracheostomy-dependent children experience significant caregiver burden.[6] Future studies on the improvement of caregiver education at discharge and reducing caregiver burden may be important in decreasing

Table 7. Patient Characteristics by 3-Year Decannulation Rates

Characteristic	Overall (n = 46)[a]	Not decannulated (n = 28)[a]	Decannulated (n = 18)[a]
Comorbid conditions[b]	4.00 (2.75, 5.00)	4.00 (3.00, 4.75)	4.00 (2.00, 5.00)
PMA at discharge	88 (69, 108)	92 (73, 106)	77 (63, 115)
Severe BPD	31 (67%)	20 (71%)	11 (61%)
COI[c]			
Very high	7 (16%)	3 (11%)	4 (24%)
High	6 (14%)	3 (11%)	3 (18%)
Moderate	5 (11%)	3 (11%)	2 (12%)
Low	12 (27%)	11 (41%)	1 (6%)
Very low	14 (32%)	7 (26%)	7 (41%)
Early adverse event			
0	26 (57%)	15 (54%)	11 (61%)
1	15 (33%)	9 (32%)	6 (33%)
2	4 (9%)	3 (11%)	1 (6%)
3	1 (2%)	1 (4%)	0 (0%)
Late adverse events[d]	4.50 (3.00, 5.75)	5.00 (3.00, 5.00)	4.00 (3.00, 6.00)
ENT OR trips	6.50 (3.00, 10.0)	7.50 (3.25, 10.0)	6.00 (2.75, 10.5)

Abbreviations: BPD, bronchopulmonary dysplasia; ENT OR, ear, nose, and throat operating room; IQR, interquartile range; IUGR, intrauterine growth retardation; NEC, necrotizing enterocolitis; PMA, postmenstrual age; SGA, small for gestational age.
[a]Median (IQR); n (%).
[b]Comorbid conditions: IUGR, SGA, NEC, pulmonary hypertension, cardiac condition, neurologic condition, metabolic condition, and genetic syndromes.
[c]Childhood opportunity index (n = 44).
[d]n = 44.

rates of tracheostomy complications and readmissions in this population. COI lacks specificity and is based on average zip code data, rather than actual patient data, which limits the specificity of socioeconomic status for this study. Further research regarding other social determinants of health and their effect on tracheostomy outcomes is needed.

Patients in this study received tracheostomy at a median 56 weeks PMA. This is a later age at placement than some other institutions have reported, with a multi-institutional study showing a median of 48 weeks PMA at tracheostomy placement.[7] The decision to proceed with tracheostomy is quite challenging, especially given the absence of evidence to support tracheostomy criteria and timing.[9] Our center is a national referral center for BPD care, known for a low tracheostomy rate, and many families travel here with the hope of avoiding tracheostomy. Our clinical practice offers multiple extubation attempts for appropriate patients in an attempt to avoid tracheostomy if that is the family's goal. We feel comfortable with this later time to tracheostomy and supporting older patients on non-invasive positive pressure for longer periods of time, as long as they are functionally doing well with good neurodevelopmental and growth outcomes. Outcomes for our development and growth-focused BPD program support this practice.[10]

With a paucity of guidelines on optimal timing of tracheostomy placement in children with BPD, it is difficult to know the ramifications of later tracheostomy. Even in critically ill adult patients, the evidence on early tracheostomy placement after initiation of mechanical ventilation is mixed with no overall effect on mortality.[11] General considerations for age at tracheostomy placement should include the risks inherent with tracheostomy, duration of neurosedative medications, neurodevelopment, and respiratory stability of the patient.

Although later tracheostomy placement may allow patients more time for ventilator weaning and possible extubation, associations of later tracheostomy placement could include higher rates of vocal cord immobility, airway stenosis, and malacia. Large airway malacia in this cohort was common with 35% and 33% of patients having tracheomalacia or bronchomalacia, respectively. These rates are comparable to published rates of malacia in ventilator-dependent children as well as infants with BPD who undergo bronchoscopy.[12,13] There does not appear to be an increase in malacia in tracheostomy-dependent children with BPD. Prospective studies examining the rates of malacia in this group could be more useful in determining malacia rates before and after tracheostomy.

The mortality in our cohort was 24% of all patients dying before primary discharge (30% overall mortality of patients for the study duration). We suspect this mortality rate reflects the nature of our BPD program, where we care for patients who are referred to our program at later ages with severe BPD and multisystem involvement. Often, these patients are referred to our program after the referring team has exceeded treatment options. As reflected in the cause of death listed in Supplemental Table S2, available online, 65% of these cases had compassionate withdrawal of care by the family. We suspect the severe multisystem involvement and

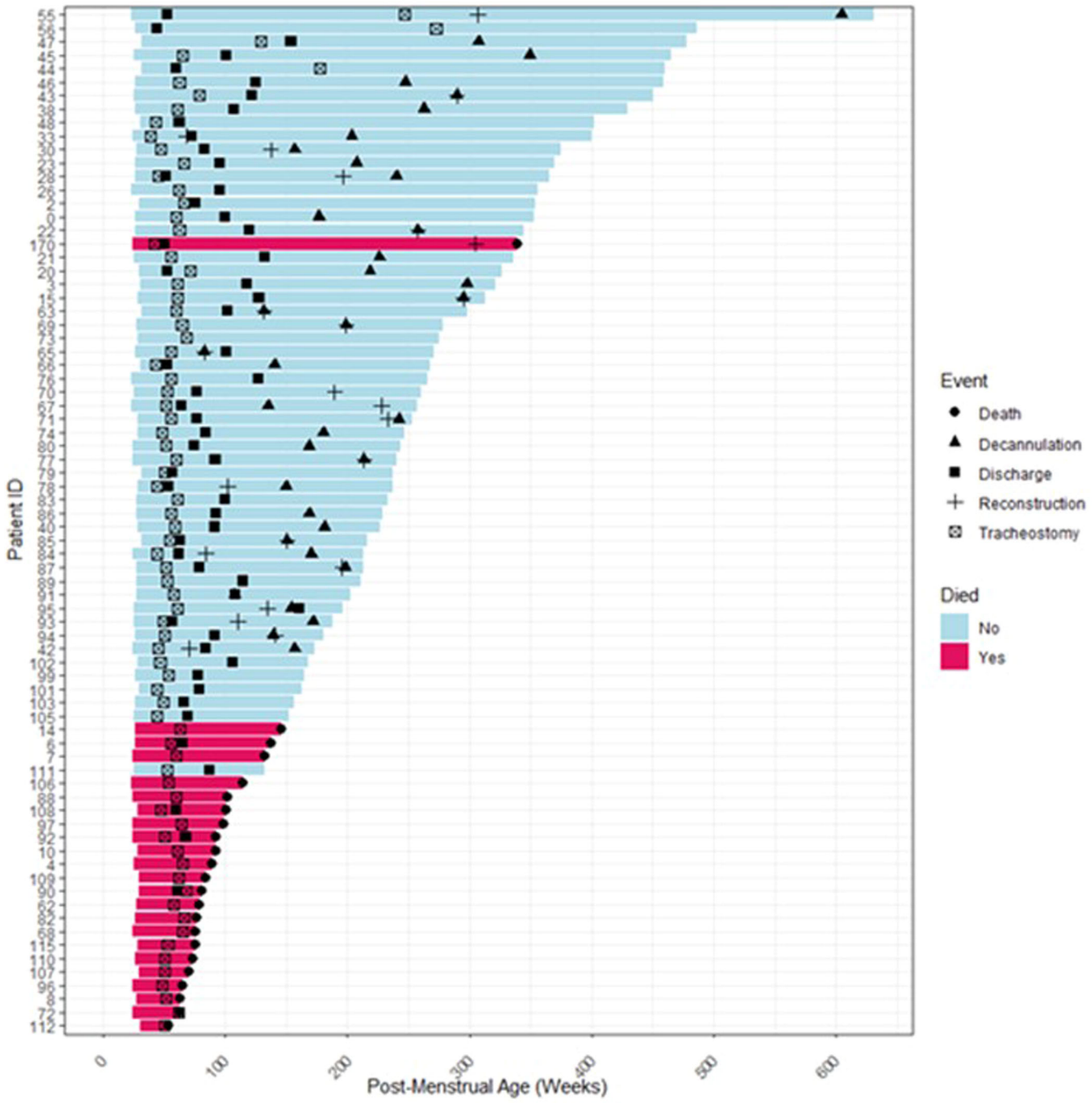

Figure 2. Swimmer's plot of tracheostomy outcomes of 76 patients with bronchopulmonary dysplasia who received tracheostomy between 2016 and 2022.

comorbidities seen in these patients also impact survival. Additionally, 12% of these patients died from non-BPD causes, including hepatoblastoma and sepsis.

This mortality rate is comparable to a similar-sized study on patients with severe BPD and tracheostomy in which 20% of patients died before primary discharge.[14] Tracheostomy-related mortality was low in our study with only one patient (1.7%) dying from accidental decannulation, despite 38% of surviving patients having a documented accidental decannulation event. Our reported tracheostomy-related mortality rate was lower than rates reported in studies on children with tracheostomies, with one study reporting 27% of mortalities from tracheostomy-related causes.[15] This may be related to caregiver education including routine and emergency tracheostomy care that is standard at our institution.

Although the tracheostomy-related mortality was low in this group, the prevalence of tracheostomy complications was high, with 50% of patients having some adverse event in the first postoperative week and 94% of surviving

patients having one or more adverse event over the follow-up period. These complication rates are much greater than published rates of late complications reported as 38.8% and 43%.[16,17] Notably, accidental decannulation occurred in more than a third of surviving patients, and 7% of discharged patients were readmitted for accidental decannulation. Other retrospective studies in tracheostomy-dependent children have reported lower rates of accidental decannulation between 0.7% and 20%.[18] However, a more recent prospective study showed that 67% of admitted tracheostomy-dependent children had at least one accidental decannulation with very few requiring serious intervention, conveying accidental decannulation as a common occurrence.[18] Comorbid conditions, patient age, and socioeconomic status may play a role in the rate of accidental decannulation observed in this cohort, but further studies are needed on how to mitigate accidental decannulation.

The most common early and late tracheostomy-related adverse event was bacterial tracheitis, with 81% of surviving patients having had at least one episode. Additionally, tracheitis was the most common reason for readmission both in the first 30 days after primary discharge and 2 years after discharge, surpassing BPD exacerbations and viral illnesses. Although rates of tracheitis are high in mechanically ventilated patients, the incidence in children with severe BPD appears to be higher than ventilated pediatric patients (81% vs 6.92%-7.3%),[19,20] possibly related to prolonged hospitalization and steroid administration in this group. To our knowledge, the elevated incidence of tracheitis in this group has not been highlighted in prior studies. Improved management of tracheitis in the future could reduce the number of hospitalizations in this cohort, but evidence-based preventative measures are limited, and diagnostic and treatment guidelines are evolving.[19]

Airway stenosis rates were high in this cohort, with 69% of surviving patients having laryngotracheal stenosis (subglottic or tracheal) following tracheostomy placement. These rates appear to be higher compared to the literature on airway stenosis, with one study finding 21% of pediatric patients who underwent tracheostomy developed SGS and another finding that 8.8% of adults developed tracheal stenosis.[21,22] However, we did not look specifically at preoperative or operative findings to determine if airway stenosis was present before tracheostomy placement. Further studies are needed to investigate the effect of timing of tracheostomy placement in patients with BPD as well as the interplay between large airway changes in BPD and airway stenosis.

Open and endoscopic airway procedures were common in our cohort. Almost half of all surviving patients in our cohort received open airway reconstructive surgery, with laryngotracheal reconstruction being the most common open procedure. To our knowledge, the high rate of open airway reconstructive surgery has not been documented in this group. The high rate of airway stenosis and subsequent airway reconstruction is not surprising, however, as patients in this group were ventilator-dependent and tracheostomy-dependent for a prolonged period. Additionally, tracheostomy-dependent BPD patients are often steroid-dependent, have recurrent infections, and have difficulties feeding, leading to poorer wound healing and potentially higher risk for posttracheostomy airway scar formation and stenosis.

The overall decannulation rate for surviving patients of 68% in this cohort was high compared to similar studies, which reported 32% to 50%.[7,23,24] Time at decannulation varies in the literature from 2.25 to 4 years of age, with the patients in our cohort decannulated at a median 3.1 years, although not all patients were followed through time to deccannulation.[7,24] Overall, surgical intervention was needed in a majority of patients for decannulation: 19% of patients were decannulated with single-stage open airway reconstruction, and 47% of decannulated patients required surgical closure of a tracheocutaneous fistula.

When examining barriers to decannulation at 3 years of age, 25% of the patients in this study were working towards decannulation and were no longer oxygen-dependent, whereas the others were still ventilator-dependent or reliant on tracheostomy for secretion management. With greater follow-up time, it is possible that more patients in this group will achieve decannulation.

Study Limitations

This study is limited in several aspects. First, it is a relatively small retrospective study of heterogeneous and complex patients without direct comparisons to a control cohort of infants with BPD who were not tracheostomy-dependent. Additionally, this study was conducted at a tertiary care children's hospital with a specialized BPD unit, which could bias our population toward more complex patients, thus affecting the wider applicability of our findings. Importantly, our site performs tracheostomies in BPD infants at a later age, which may influence the airway outcomes and complications compared to other institutions. Finally, although tracheostomy placement for BPD appears to be becoming more common, the size limitations of our study prevented greater statistical analysis. Larger, multicenter studies are needed to further elucidate the overall outcomes and airway interventions of tracheostomy-dependent infants with BPD.

Conclusion

In this retrospective cohort study, we described the tracheostomy outcomes of infants with BPD, highlighting some findings previously not reported in the literature. Tracheostomy-dependent infants with BPD are a medically complex group with a significant risk of mortality, tracheostomy complications, and readmissions. Most notably in this study was the high percentage of readmissions for tracheitis and

accidental decannulation. Also notable in this cohort were the high rates of laryngotracheal stenosis and subsequent open airway reconstruction. Future studies should focus on optimal timing of tracheostomy placement, tracheitis mitigation, the progression of laryngotracheal stenosis, and subsequent timing of open airway reconstruction.

Author Contributions

Ada Cleary Sher, conception and study design, data acquisition, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Humra Shamim**, data acquisition, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Jemma Maynard**, data acquisition, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Jacob Stack**, data acquisition, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Isaac Kistler**, data analysis, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Megan McNutt**, data acquisition, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Hajera Afreen**, conception and study design, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Amy Manning**, conception and study design, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Audrey Miller**, conception and study design, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Prasanth Pattisapu**, conception and study design, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work; **Tendy Chiang**, conception and study design, data interpretation, drafting manuscript and critical revision, final approval of version to be published, agreement to be accountable for all aspects of the work.

Disclosures

Competing interests: None.
Funding source: None.

Supplemental Material

Additional supporting information is available in the online version of the article.

ORCID iD

Ada Cleary Sher http://orcid.org/0000-0002-1238-2953
Jemma Maynard http://orcid.org/0009-0008-1702-5112
Jacob Stack http://orcid.org/0009-0006-1009-9050
Isaac Kistler http://orcid.org/0009-0005-4025-1327
Megan McNutt http://orcid.org/0009-0002-2946-5656
Prasanth Pattisapu https://orcid.org/0000-0002-6857-2086

References

1. Stoll BJ, Hansen NI, Bell EF, et al. Neonatal outcomes of extremely preterm infants from the NICHD Neonatal Research Network. *Pediatrics*. 2010;126(3):443-456. doi:10.1542/peds.2009-2959
2. Dankhara N, Holla I, Ramarao S, Kalikkot Thekkeveedu R. Bronchopulmonary dysplasia: pathogenesis and pathophysiology. *J Clin Med*. 2023;12(13):4207. doi:10.3390/jcm12134207
3. Kalikkot Thekkeveedu R, Guaman MC, Shivanna B. Bronchopulmonary dysplasia: a review of pathogenesis and pathophysiology. *Respir Med*. 2017;132:170-177. doi:10.1016/j.rmed.2017.10.014
4. Guaman MC, Pishevar N, Abman SH, et al. Invasive mechanical ventilation at 36 weeks post-menstrual age, adverse outcomes with a comparison of recent definitions of bronchopulmonary dysplasia. *J Perinatol*. 2021;41(8):1936-1942. doi:10.1038/s41372-021-01102-w
5. Jensen EA, Dysart K, Gantz MG, et al. The diagnosis of bronchopulmonary dysplasia in very preterm infants an evidence-based approach. *Am J Respir Crit Care Med*. 2019;200(6):751-759. doi:10.1164/rccm.201812-2348OC
6. Hartnick CJ, Bissell C, Parsons SK. The impact of pediatric tracheotomy on parental caregiver burden and health status. *Arch Otolaryngol Head Neck Surg*. 2003;129(10):1065. doi:10.1001/archotol.129.10.1065
7. Manimtim WM, Agarwal A, Alexiou S, et al. Respiratory outcomes for ventilator-dependent children with bronchopulmonary dysplasia. *Pediatrics*. 2023;151(5):e2022060651. doi:10.1542/peds.2022-060651
8. Smith MA, Steurer MA, Mahendra M, Zinter MS, Keller RL. Sociodemographic factors associated with tracheostomy and mortality in bronchopulmonary dysplasia. *Pediatr Pulmonol*. 2023;58(4):1237-1246. doi:10.1002/ppul.26328
9. Miller AN, Shepherd EG, Manning A, et al. Tracheostomy in severe bronchopulmonary dysplasia—how to decide in the absence of evidence. *Biomedicines*. 2023;11(9):2572. doi:10.3390/biomedicines11092572
10. Kielt MJ, Logan JW, Backes CH, Reber KM, Nelin LD, Shepherd EG. In-hospital outcomes of late referrals for established bronchopulmonary dysplasia. *J Perinatol*. 2021;41(8):1972-1982. doi:10.1038/s41372-021-01041-6
11. Bice T, Nelson J, Carson S. To trach or not to trach: uncertainty in the care of the chronically critically ill. *Semin Respir Crit Care Med*. 2015;36(6):851-858. doi:10.1055/s-0035-1564872
12. Manimtim WM, Rivard DC, Sherman AK, et al. Tracheobronchomalacia diagnosed by tracheobronchography in ventilator-dependent infants. *Pediatr Radiol*. 2016;46(13):1813-1821. doi:10.1007/s00247-016-3685-9
13. Hysinger EB, Friedman NL, Padula MA, et al. Tracheobronchomalacia is associated with increased morbidity in bronchopulmonary dysplasia. *Ann Am Thorac Soc*. 2017;14(9):1428-1435. doi:10.1513/AnnalsATS.201702-178OC

14. Upadhyay K, Vallarino DA, Talati AJ. Outcomes of neonates with tracheostomy secondary to bronchopulmonary dysplasia. *BMC Pediatr*. 2020;20(1):414. doi:10.1186/s12887-020-02324-1
15. Hebbar KB, Kasi AS, Vielkind M, et al. Mortality and outcomes of pediatric tracheostomy dependent patients. *Front Pediatr*. 2021;9:661512. doi:10.3389/fped.2021.661512
16. Mahadevan M, Barber C, Salkeld L, Douglas G, Mills N. Pediatric tracheotomy: 17 year review. *Int J Pediatr Otorhinolaryngol*. 2007;71(12):1829-1835. doi:10.1016/j.ijporl.2007.08.007
17. Watters K, O'Neill M, Zhu H, Graham RJ, Hall M, Berry J. Two-year mortality, complications, and healthcare use in children with Medicaid following tracheostomy. *Laryngoscope*. 2016;126(11):2611-2617. doi:10.1002/lary.25972
18. Wynings EM, Breslin N, Brooks RL, et al. Accidental tracheostomy decannulations in children—a prospective cohort study of inpatients. *Laryngoscope*. 2023;133(4):963-969. doi:10.1002/lary.30250
19. Ormsby J, Conrad P, Blumenthal J, et al. Practice improvement for standardized evaluation and management of acute tracheitis in mechanically ventilated children. *Pediatr Qual Saf*. 2021;6(1):e368. doi:10.1097/pq9.0000000000000368
20. Beardsley AL, Rigby MR, Bogue TL, Nitu ME, Benneyworth BD. The incidence of ventilator-associated infections in children determined using bronchoalveolar lavage. *Glob Pediatr Health*. 2015;2:2333794X15580771. doi:10.1177/2333794x15580771
21. Pinzas LA, Bedwell JR, Ongkasuwan J. Glottic and subglottic injury and development of pediatric airway stenosis. *Otolaryngol Head Neck Surg*. 2023;168(3):469-477. doi:10.1177/01945998221100829
22. James P, Parmar S, Hussain K, Praveen P. Tracheal stenosis after tracheostomy. *Br J Oral Maxillofac Surg*. 2021;59(1):82-85. doi:10.1016/j.bjoms.2020.08.036
23. Akangire G, Manimtim W. Tracheostomy in infants with severe bronchopulmonary dysplasia: a review. *Front Pediatr*. 2023;10:10. doi:10.3389/fped.2022.1066367
24. Akangire G, Lachica C, Noel-MacDonnell J, et al. Outcomes of infants with severe bronchopulmonary dysplasia who received tracheostomy and home ventilation. *Pediatr Pulmonol*. 2023;58(3):753-762. doi:10.1002/ppul.26248