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Positive Airway Pressure Ventilation & Complications in Pediatric Tracheocutaneous Fistula Repair

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Abstract

Objectives: Surgical repair of persistent tracheocutaneous fistula in children may be complicated by tracheal air leak with resultant subcutaneous emphysema, pneumomediastinum, and/or pneumothorax. We first sought to identify clinical risk factors for post-operative complications after primary repair of persistent tracheocutaneous fistula in children. Second, the type and frequency of complications in patients administered positive airway pressure ventilation (e.g. bag-valve mask ventilation, CPAP, or BiPAP) post-operatively was determined and compared to a control population.

Methods: This was a retrospective investigation of all pediatric patients (n = 108) undergoing surgical repair of persistent tracheocutaneous fistula from January 2000 – April 2016 at a tertiary, academic referral center. Type and frequency of post-operative complications were compared among patients administered positive airway pressure ventilation post-operatively vs. those not.

Results: Of 108 pediatric patients, complications after tracheocutaneous fistula repair occurred in 22 (20.4 %) patients. These included symptoms of respiratory distress requiring intervention (e.g. supplemental O_2, racemic epinephrine, intubation), subcutaneous emphysema, pneumomediastinum, and/or pneumothorax, bleeding, wound infection, and readmission. Frequency of all post-operative complications were significantly higher in patients administered positive airway pressure ventilation vs. those not (50.0 % vs. 16.7 %, $p = 0.015$), as were rates of subcutaneous emphysema, pneumomediastinum, and/or pneumothorax (33.3 % vs. 4.2 %, $p = 0.005$).

Conclusions: Positive airway pressure ventilation after primary repair of persistent tracheocutaneous fistula in children may increase risk of serious respiratory complications. In practice, we advocate for avoidance of bag-valve mask ventilation and caution when utilizing CPAP or BiPAP post-operatively in these patients.

Keywords: Pediatric, tracheocutaneous fistula, complications, positive airway pressure

Level of Evidence: 4

Introduction

Common indications for tracheostomy in the pediatric population include prolonged ventilator dependence, neuromuscular disease, and congenital airway anomalies.[1-3] These chronic diseases often necessitate prolonged duration of tracheostomy. Longer duration of tracheostomy (>12 months) and younger age at time of tracheostomy (<6 months) is associated with significantly increased risk of persistent tracheocutaneous fistula (TCF) after decannulation.[4] While published estimates vary, as many as 40 % of children will have persistent TCF after decannulation if they had a tracheostomy for greater than one year.[5,6]

Persistent TCF causes impaired cough, altered phonation, egress of mucus with skin irritation, and social discomfort.[7] Additionally, swimming must be avoided due to inability to protect the airway. This has significant impact on quality of life for some patients. Two common surgical techniques are used to treat TCF; excision of the fistulous tract with primary multi-layered closure or tract excision with healing by secondary intention.[8] Both methods offer high success rates for TCF closure and have no differences in post-operative complication rates.[4,7] Minor complications after these operations include wound infection, dehiscence, recurrence of the TCF, unsightly scar, and need for revision surgery.[9,10] Major complications include subcutaneous emphysema, pneumomediastinum, and pneumothorax. These life-threatening complications are caused by air escape into the subcutaneous tissues of the neck, mediastinum, and thorax through elevated airway pressures.[11]

Noninvasive positive airway pressure (PAP) ventilation, in the form of bag-valve mask ventilation, continuous positive airway pressure (CPAP), or bilevel positive airway pressure (BiPAP), may precipitate these major complications post-operatively. Pediatric patients with tracheostomy frequently have comorbid respiratory issues and some require nocturnal or continuous PAP ventilation but otherwise qualify for decannulation. This poses unique clinical challenges for postoperative management following TCF closure.

We present a retrospective cohort study of pediatric patients who underwent surgical repair of persistent TCF at our institution from January 2000 to April 2016. Our primary objective was to identify risk factors for post-operative complication following TCF repair, with particular focus on assessing the effect of postoperative PAP following TCF repair. To our knowledge, this is the first study evaluating the effect of PAP use after TCF repair on postoperative complications.

Materials and Methods

All pediatric patients under age 18 at the time of surgery who underwent TCF repair at a tertiary, academic referral center from January 2000 – April 2017 were included for retrospective chart review (n = 108). A cohort discovery tool, DataDirect, was used to identify subjects for inclusion in this study.[12] Query criteria included patients under the age of 18 at the time of surgery, those with a diagnosis of TCF, and those undergoing TCF repair identified in the medical record by CPT code 31825.

Data extracted from the medical record included: patient demographics, medical comorbidities, indication for tracheotomy, tracheotomy technique (i.e. normal vs. permanent/fenestrated), duration of tracheostomy cannulation, interval from decannulation to TCF repair, use of PAP post-operatively, and post-operative complications. A normal tracheotomy technique consisted of a vertical tracheal incision between the third and fourth tracheal rings followed by placement of stay sutures. A permanent/fenestrated tracheotomy consisted of formalization of the tracheostoma by sewing an inferiorly-based tracheal flap to the skin or subcutaneous tissues. All patients in our study had persistent TCF repaired primarily. The procedure starts with diagnostic laryngoscopy and bronchoscopy to verify adequate airway for TCF repair. The repair consists of an elliptical skin incision followed by dissection of the tract carried down to the level of the trachea. The fistulous tract is excised and the tracheal window is closed primarily using interrupted Vicryl sutures. Valsalva maneuver is used to confirm no intraoperative air leak. A passive, sterile rubber band is placed deep to the sternohyoid and sternothyroid muscles, and the muscles and skin are approximated loosely.

For this study, we included the following as post-operative complications: symptoms/signs of respiratory distress meriting medical or surgical intervention, subcutaneous emphysema, pneumomediastinum, and/or pneumothorax, incisional bleeding, wound infection (i.e. superficial erythema and/or purulence treated with oral antibiotics), and readmission. All patients documented to have increased work-of-breathing, retractions, stridor, dyspnea, or hypoxia meriting medical or surgical intervention, but without evidence of subcutaneous emphysema, pneumomediastinum, and/or pneumothorax post-operatively were categorized as having complication of respiratory distress.

Student's t-test and Fisher's exact test were used to compare continuous and categorical variables, respectively, among patients who experienced post-operative complications vs. those who had no complications after TCF repair (two-tailed, $\alpha = 0.05$). Statistical analysis was performed using Stata software (StataCorp, College Station, TX). This study was approved by the University of Michigan Institutional Review Board with waiver of informed consent.

Results

During the study period, a total of 108 pediatric patients underwent surgical repair of persistent TCF. Demographic characteristics of our cohort are depicted in **Table 1**. The original tracheotomy technique was standard technique in 96 (88.9 %) patients, permanent technique in nine (8.3 %), and unknown for three (2.8 %). Within our entire pediatric cohort, mean (SD) duration of tracheostomy cannulation was 40.4 (± 29.6) months while the mean (SD) time interval from tracheostomy decannulation to TCF repair was 11.6 (± 17.4) months.

Complications from TCF repair occurred in 22 (20.4 %) patients. These complications included post-operative respiratory distress requiring intervention (e.g. supplemental O_2, racemic epinephrine, intubation) in 9 (8.3 %) patients (**Table 2**), subcutaneous emphysema, pneumomediastinum, and/or pneumothorax in 8 (7.4 %), bleeding in 2 (1.9 %), wound infection in 2 (1.9 %), and readmission in 1 (0.9 %). To identify potential risk-factors for complications from TCF repair, we compared comorbidities, tracheotomy technique, duration of tracheostomy cannulation, tracheostomy size at decannulation, interval from decannulation to TCF repair, and use of positive-airway pressure (PAP) post-operatively among pediatric patients with and without post-operative complications (**Table 3**). A significantly higher proportion of patients with complications from TCF repair had undergone a permanent/fenestrated tracheotomy (18.2 % vs. 5.8 % of patients with no complications, $p = 0.021$). Importantly, pediatric patients with complications from TCF repair were more frequently administered PAP ventilation (i.e. bag-valve mask ventilation, CPAP, or BiPAP) in the post-operative period (27.3 % vs. 7.0 % of patients with no complications, $p = 0.015$).

The most common complication in patients administered PAP post-operatively was subcutaneous emphysema, pneumomediastinum, and/or pneumothorax (**Table 4**). To further investigate the relationship between post-operative PAP use and complications, we stratified this subset of patients into those with planned PAP use (i.e. CPAP or BiPAP for pre-existing obstructive sleep apnea or central apnea) and those with unplanned PAP use (i.e. bag-mask ventilation in PACU) post-operatively. This analysis revealed a significantly increased risk of complications, namely subcutaneous emphysema, pneumomediastinum, and/or pneumothorax among patients with unplanned post-operative PAP use only. The complication rate in the planned PAP group was similar to the complication rate among patients with no postoperative PAP use (14.3% for planned PAP, 16.7% no PAP) (**Table 4**).

Within our entire cohort, the mean (SD) post-operative ICU and hospital stays were 0.96 (± 1.47) and 1.67 (± 1.56) days, respectively. Patients administered PAP post-operatively had significantly longer ICU stays (2.0 ± 2.09 vs. 0.83 ± 1.34 days, $p = 0.009$) and hospital stays (2.50 ± 1.83 vs. 1.56 ± 1.50 days, $p = 0.049$) compared to patients not administered PAP post-operatively.

Discussion

This is the first study examining the safety and potential clinical sequelae of PAP ventilation after surgical repair of persistent TCF in a large pediatric population. Our results suggest that post-operative PAP use, particularly unplanned PAP use in the form of bag-valve mask ventilation for respiratory symptoms immediately post-operatively, increase rates of subcutaneous emphysema, pneumomediastinum, and/or pneumothorax and prolong total ICU and hospital stay.

All patients in our study underwent TCF closure via excision of the fistulous tract with primary multi-layered closure over a passive rubber band drain. This surgical technique affords immediate resolution of the TCF, requires minimal post-operative care by patient and parents, and may offer superior rates of successful closure without TCF recurrence or need for scar revision.[13] However, the main concern with primary closure of TCF is air leaking from the airway and subcutaneous trapping of this air, causing subcutaneous emphysema, pneumomediastinum, and/or pneumothorax and potentially necessitating urgent airway interventions or chest tube placement.[14-16] Intraoperative air leak check, loose closure of superficial tissues, and placement of a passive rubber band drain are utilized strategies to decrease the risk of subcutaneous air accumulation and progression at our institution. However, whether drains or specific types thereof indeed reduce post-operative complications after TCF repair has yet to be shown objectively.

Excision of the fistulous tract with healing by secondary intention is used by some surgeons to minimize the risk for subcutaneous emphysema, pneumomediastinum, and pneumothorax post-operatively.[9] A recent systematic review and meta-analysis of eight studies comprising 259 pediatric patients undergoing TCF repair reported an increased rate of these major respiratory complications among patients treated with primary closure compared to fistulectomy and healing by secondary intention (8.5 % vs. 0.0 %, p = 0.038).[17] Conversely, a larger meta-analysis found no difference in rates of all complications and subcutaneous emphysema/pneumothorax in pediatric patients undergoing primary or secondary closure of persistent TCF (10 % vs. 9.9 %, p = 0.551, 3.8 % vs. 3.8 %, p = 0.512, respectively).[4]

The rate of all post-operative complications in our pediatric cohort undergoing primary closure of persistent TCF was 20.4 %, higher than other published reports.[4,7,9,14,17,18] We suspect that this difference is partially explained by variation in classification and inclusion of postoperative complications from study to study. Our rates of bleeding, surgical-site infection, and subcutaneous emphysema, pneumomediastinum, and pneumothorax were quite similar to published reports. However, we purposely included the complication of "symptoms/signs of respiratory distress meriting medical or surgical intervention," whereas other studies did not. We did so to elaborate on rates and complex management of respiratory complications distinct from subcutaneous emphysema, pneumomediastinum, and pneumothorax that may follow primary TCF repair.

The nine patients experiencing respiratory distress post-operatively did not significantly differ from the remaining patients in number and nature of comorbidities (**Table 2**). These patients were often reintubated and admitted to the PICU for further management. Of note, four of these patients had repeat direct laryngoscopies with excision of suprastomal granulation tissue that likely contributed to resolution of their symptoms of respiratory distress. In retrospect, this non-obstructing granulation was noted and left in place at the time of TCF repair in each of these patients. Perhaps complete excision of suprastomal granulation tissue is warranted at time of TCF repair to prevent this from contributing to post-operative respiratory distress.

Identification of demographic and/or clinical features predictive of complications after surgical repair of persistent TCF would certainly be valuable in pre-op assessment of surgical candidacy. Unfortunately, our study failed to identify any tracheostomy variables, airway characteristics or medical comorbidities more frequently present in children experiencing complications after surgery (**Table 2**). Thus, assessment of candidacy for surgical repair of persistent TCF should be left to the discretion of experienced pediatric otolaryngologists.

We propose that PAP ventilation may be an iatrogenic precipitant of tracheal air leak associated with serious respiratory complications after TCF repair.[19,20] Importantly, the 12 pediatric patients administered PAP post-operatively segregated into two distinct cohorts. In the “unplanned PAP” group, patients were treated with noninvasive ventilation for a number of indications including; stridor, hypoxia, increased work of breathing and, dyspnea in the PACU. All patients in this group subsequently developed subcutaneous emphysema or pneumothorax requiring re-intubation or chest tube placement. In the “planned PAP” group, patients with comorbid OSA or central apnea were administered nocturnal CPAP or BiPAP to maintain saturations in accordance with their home pulmonary regimen, and were monitored closely in-hospital for development of subcutaneous air.

Timing of post-operative PAP use may be a critical factor influencing risk for tracheal air leak. Perhaps the longer time interval from TCF repair to nocturnal PAP use, as in the “planned PAP” group, is protective and affords time for initial healing and wound stabilization. Alternatively, bag-valve mask ventilation may deliver higher pressures and contribute to air escape. However, we cannot definitively conclude that unplanned PAP use caused subcutaneous emphysema, pneumomediastinum, and pneumothorax in this retrospective review. It is certainly possible that these patients were exhibiting early signs and symptoms of these complications, and bag-valve mask ventilation simply worsened their clinical status. Importantly however, each patient in our cohort experiencing subcutaneous emphysema, pneumomediastinum, and/or pneumothorax were reexamined endoscopically either immediately post-operatively or later during follow-up. Recurrence of the original indication for tracheostomy (e.g. subglottic stenosis) was not seen in any of our patients, lending support to our hypothesis that PAP ventilation directly caused tracheal air leak.

Nevertheless, our results suggest that PAP use in the immediate post-operative period after primary closure of persistent TCF should be avoided. Furthermore, clear communication with anesthesia and nursing staff in the PACU and PICU regarding risks of PAP ventilation in these patients is essential. We advocate for prompt tracheal re-intubation when signs and symptoms of respiratory distress develop post-operatively. Alternatively, it may be reasonable to plan for a temporary, 24-hour period of intubation after primary TCF repair in "high-risk" children with significant comorbidities to reduce the need for PAP and allow for initial healing of the tracheal closure. Further, we recommend judicious use of nocturnal CPAP or BiPAP in patients with comorbid OSA or central apnea and close monitoring for subcutaneous air in-hospital by all medical staff.

Limitations of this study include retrospective study design, which limits conclusions of causality between treatments and complications mentioned above. For some cases, it was impossible to determine whether PAP use preceded respiratory symptoms and airway leak or coincided with this complication. Also, we found an increased risk of post-operative complications in patients who underwent a permanent tracheotomy technique. We hypothesize that this represents a selection bias rather than a true risk-factor, as different patient populations will be selected for different procedures such as permanent tracheostomy technique.

Conclusions

In a large pediatric population undergoing primary closure of persistent TCF, we found that post-operative PAP, especially mask ventilation in the PACU, was associated with an increased risk of serious complications, such as subcutaneous emphysema, pneumomediastinum, and pneumothorax. We advocate for avoidance of bag-valve mask ventilation and caution with close monitoring when utilizing CPAP or BiPAP post-operatively in patients undergoing primary repair of TCF.

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TABLE I. Characteristics of Patients (n = 108) Undergoing TCF Repair†

Sex	
Male	64 (59.3 %)
Female	44 (40.7 %)
Tracheostomy Indication	
Ventilator Requirement	64 (59.3 %)
Upper Airway Obstruction	22 (20.4 %)
Subglottic Stenosis	16 (14.7 %)
Glottic Obstruction	4 (3.7 %)
Neurologic Disease	2 (1.9 %)
Age at Tracheotomy (months)	13.6 (± 34.6)

† Data presented as n (%) or mean (SD)

TABLE II. Description of Patients Experiencing Complication of Post-Operative Respiratory Distress without Evidence of Subcutaneous Emphysema, Pneumomediastinum, and/or Pneumothorax (n = 9)

Patient	Setting	Symptoms/Signs	Intervention(s)
1	PACU	Retractions, Stridor	Direct laryngoscopy & bronchoscopy, excision of suprastomal granulation tissue
2	Home	Dysphonia, Stridor	Direct laryngoscopy & bronchoscopy, excision of suprastomal granulation tissue
3	PACU	Hypoxia	Nebulized ipratropium, fluticasone, admission to PICU
4	Home	Hypoxia, Stridor	Readmission, IV dexamethasone, direct laryngoscopy & bronchoscopy, excision of suprastomal granulation tissue, replacement of tracheostomy tube
5	PACU	Hypoxia, Retractions, Stridor, Cough, Dyspnea, Tachypnea	Racemic epinephrine, albuterol, O_2, direct laryngoscopy & bronchoscopy, reintubation
6	Inpatient Floor	Hypoxia, Retractions, Wheezing	Nebulized fluticasone, albuterol
7	PACU	Hypoxia, Stridor	Nebulized fluticasone, racemic epinephrine, dexamethasone, O_2, reintubation, admission to PICU
8	PACU	Hypoxia, Apnea	IV dexamethasone, reintubation, admission to PICU
9	PACU	Hypoxia, Stridor	Direct laryngoscopy & bronchoscopy, excision of suprastomal

	granulation tissue, IV dexamethasone, reintubation, admission to PICU

TABLE III. Comparison of Clinical Variables in Patients with Complications from TCF Repair vs. Those with No Complications†

	No Complication(s) (n = 86)	Complication(s) (n = 22)	*p* Value
Comorbidities			
Pulmonary Disease	51 (59.3 %)	12 (57.1 %)	0.686
Neurologic Disease	33 (38.4 %)	10 (45.5 %)	0.545
Prematurity	29 (33.7 %)	8 (36.4 %)	0.816
Cardiac Disease	18 (20.9 %)	7 (31.8 %)	0.280
Craniofacial Syndrome	18 (20.9 %)	3 (13.6 %)	0.331
Obstructive Sleep Apnea	13 (15.1 %)	4 (18.2 %)	0.472
Central Apnea	8 (9.3 %)	1 (4.6 %)	0.416
Morbid Obesity	3 (3.5 %)	0 (0 %)	0.501
Tracheotomy Technique‡			***0.021***

Normal	80 (93.0 %)	16 (72.7 %)	
Permanent/Fenestrated	5 (5.8 %)	4 (18.2 %)	
Duration of Trach Cannulation (months)	40.9 (± 31.0)	38.4 (± 24.4)	0.349
Trach Inner Diameter (ID) at Decannulation (cm)	3.8 (± 0.65)	3.9 (± 0.63)	0.616
Decannulation to TCF Repair Interval (Months)	12.1 (± 18.3)	9.8 (± 13.6)	0.533
Post-Operative Positive Airway Pressure (PAP) Use	6 (7.0 %)	6 (27.3 %)	***0.015***

† *Data presented as n (%) or mean (SD)*

‡ *Tracheotomy technique was unknown/undocumented in 1 patient with no complications and 2 with complications from TCF repair*

TABLE IV. Positive Airway Pressure (PAP) and Post-Operative Complications in Pediatric TCF Repair†

	No PAP (n = 96)	**All PAP (n = 12)**	***p* Value**	**Planned PAP‡ (n = 7)**	***p* Value**	**Unplanned PAP§ (n = 5)**	***p* Value**
Any Complication	16 (16.7 %)	6 (50.0 %)	***0.015***				
Subcutaneous Emphysema,	4 (4.2 %)	4 (33.3 %)	***0.005***				

Pneumomediastinum, and/or Pneumothorax				
Any Complication	1 (14.3 %)	1.000	5 (100.0 %)	<***0.000***
Subcutaneous Emphysema, Pneumomediastinum, and/or Pneumothorax	1 (14.3 %)	0.426	3 (60.0 %)	***0.003***

† *Data presented as n (%) or mean (SD)*
‡ *CPAP or BiPAP for patients with comorbid obstructive sleep apnea (OSA) or central sleep apnea (CSA)*
§ *Bag-valve mask ventilation for post-operative respiratory distress*