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Airway findings in children with tracheostomies: When is diagnostic bronchoscopy and laryngoscopy indicated?

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Abstract

Objectives: To determine the utility of diagnostic laryngoscopy and bronchoscopy in children with tracheostomies and to describe the incidence of airway findings.

Methods: Retrospective cohort study examining children with tracheostomies who underwent direct laryngoscopy and bronchoscopy (DLB) at Duke University Hospital between 2008 and 2016.

Results: A total of 81 patients who underwent 114 bronchoscopies met inclusion criteria. The median time between tracheostomy and first DLB was 13 months (IQR 4.25-14.75). Sixty-six patients were diagnosed with findings on DLB (81.5%). Suprastomal granulation tissue was the most common complication (59.0%), followed by increased tracheal secretions (23%), stomal and peristomal granulation (13.2%), tracheal ulcer (3.3%), and suprastomal collapse (1.6%). The proportion of patients with airway findings who underwent endoscopy >6 months post-tracheostomy was higher than those <6 months post-tracheostomy, although this did not reach statistical significance (90.6% vs. 75.5%, $p=0.087$). However, when examining tracheostomy-related findings, the proportion of patients with airway findings who underwent DLB >6 months post-tracheostomy (61%) compared to <6 months post-tracheostomy (36%) was significantly different ($p = 0.026$). Patients who were symptomatic before bronchoscopy were more likely to have positive findings (91.9% vs. 72.7%, $p=0.027$) and patients were more likely to be symptomatic if they had DLB >6 months after tracheostomy versus <6 months after tracheostomy (68.8% vs. 30.6%; $p<0.001$).

Conclusion: The high incidence of airway findings, especially tracheostomy-related findings, noted on DLB supports the continued use of airway endoscopies in children post-tracheostomy.

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Timing of DLB may play a role in determining utility with evaluation and symptomatic patients should be more closely monitored as they demonstrate higher rates of airway findings.

Keywords

bronchoscopy; tracheostomy; laryngoscopy; monitoring; surveillance; airway lesion

1. Introduction

Despite advancements in care, pediatric tracheostomy remains a common surgical procedure that is associated with significant morbidity and mortality [1]. Short-term complications include bleeding, pneumothorax, and accidental decannulation whereas long-term complications include suprastomal collapse, fistula formation, and granulation tissue [2]. The variability in complications and time to occurrence has made routine airway screening with direct laryngoscopy and bronchoscopy (DLB) a common tool for monitoring and diagnosing complications after tracheostomy [3].

Currently, there are no clinical practice guidelines that address appropriate timing or indications for surveillance DLBs. This paucity is evident in the diversity of practice patterns among clinicians. A subset of pediatric otolaryngologists perform DLBs based solely on timing post-tracheostomy, most commonly at six months to one year post-operatively [4]. Others pursue DLB based on specific concerns, such as difficult tracheostomy tube changes, ventilator dependence, or bleeding [4, 5]. Still, other clinicians consider DLB when there is potential for interventions such as tracheostomy size change, decannulation, or laryngeal reconstruction [4]. Even at our institution, one pediatric otolaryngologist performed surveillance DLBs every 6 months while another only performed surveillance DLB for symptoms or concerns over tracheostomy sizing.

Two conflicting prior studies have examined the benefits of DLBs in children with tracheostomies. The high incidence of airway lesions found on screening, and subsequent need for intervention, led to the conclusion that surveillance DLBs are a useful component of post-tracheostomy care [6, 7]. At the same time, there have been recent findings that suggest a correlation between general anesthesia and irreversible neurodevelopmental impairment in pediatric populations. This has raised concern for perhaps unnecessary DLBs given the lack of explicit guidelines for frequency of endoscopic evaluation [8-11].

In light of the potential risks and benefits associated with surveillance DLBs, this study aims to determine the utility of routine airway evaluation in children with tracheostomies. We hypothesized that, while there may be a high incidence of airway findings, timing post-tracheostomy and presence of symptoms will ultimately dictate the clinical utility of endoscopic evaluations. Our study expands prior work by examining a larger subset of tracheostomy indications and a more expansive list of airway findings. Furthermore, we evaluated a more restricted historical timeframe, resulting in a more uniform protocol for comparison. We expect this study to lead to a multi-institutional collaboration that may guide the formation of practice guidelines for the use of DLBs in children with tracheostomies.

2. Material and Methods

This study was approved by the Duke University Institutional Review Board (Pro00068727). A retrospective chart review of all children with tracheostomies who underwent direct laryngoscopy and bronchoscopy (DLB) between 2008 and 2016 was conducted. A self-service, web-based clinical research query tool was used to identify patients who had tracheostomies and DLBs performed at our institution. Any patients who had tracheostomies placed at outside hospitals were excluded from analysis as the details of these procedures were not available. Additionally, patients >18 years old or those that had tracheostomy but no subsequent DLB were excluded. The initial DLB as well as any subsequent DLBs during the study period were included in the analysis. Of note, airway findings that had been noted on a previous DLB were not “recounted” as airway findings on subsequent DLBs.

Demographic, clinical, and intraoperative data was extracted from patient charts and entered into a REDCap database. Demographic variables included age and sex. Clinical data included indication for tracheostomy, tracheostomy type (e.g., cuff, cuffless, FlexTend, standard), gestational age, presence of symptoms prior to DLB, time between tracheostomy and DLB, and whether the patient was decannulated post-DLB. All DLB occurrences for an individual patient were included in our dataset.

Documented symptoms that classified a patient as “symptomatic” prior to their DLB included those that were likely tracheostomy related, those that were due to the underlying nature of the disease (i.e., reason for tracheostomy placement), and those that were independent of tracheostomy placement. Tracheostomy-related symptoms included aphonia, inability to tolerate Passy-Muir Valve or capping, bleeding, pain, difficulty with tracheostomy changes, accidental decannulation, and tracheitis. Those that were more likely due to the underlying nature of their original disease were respiratory distress, obstruction or desaturation on capped sleep studies and ventilator dependence. Symptoms independent of tracheostomy included adenotonsillar hypertrophy.

Patients that did not have any symptoms prior to their DLB were considered to be undergoing surveillance DLB.

Airway findings and any operative interventions at the time of the bronchoscopy were also recorded. Airway findings were classified as “tracheostomy-related,” persistent and independent of tracheostomy, and new and independent of tracheostomy. Tracheostomy-related findings were those that were likely a direct consequence of the tracheostomy. These included suprastomal granulation, stomal granulation tissue and peristomal granulation, tracheal ulcer, tracheal bleeding, distal tracheal granulation, suprastomal collapse, and tracheal secretions. To align with prior literature, tracheal secretions were considered to be abnormal if they were described as mucopurulent, thick, adherent or bloody in the operative report [12, 13]. Conversely, clear secretions, which are expected, were not considered abnormal findings.

Persistent and independent airway findings were those that had been present at the time of tracheostomy placement such as subglottic stenosis, subglottic cyst and tracheobronchomalacia. Finally, new and independent airway findings were those that had

not been identified at the time of tracheostomy but were likely not a direct result of the tracheostomy such as adenotonsillar hypertrophy. A “normal DLB” was one that did not yield any pertinent airway findings and was most often identified in patients who underwent tracheostomy for underlying chronic lung disease or ventilator dependence for neuromuscular disorders.

All variables were summarized using median and IQR. Proportions and frequencies were compared using χ^2 test except for the proportion of patients with symptoms before bronchoscopy who had airway findings and difference in findings depending on indication for tracheostomy, which were analyzed using Fisher’s exact test. All analyses were performed with R version 3.4.3 (Vienna, Austria).

3. Results

Between January 1st, 2008 and March 1st, 2016, 261 pediatric patients underwent tracheostomies at our hospital. A total of 81 patients who had undergone both tracheostomy placement and subsequent DLB at our institution during this timeframe met the inclusion criteria. There were 46 males and 35 females.

The most common indications for tracheostomy was ventilator dependence (64.1%), followed by TBM (37.0%), respiratory failure (17.2%), vocal cord paralysis (6.2%), congenital upper airway obstruction (3.7%), and sleep apnea (2.5%).

Among these 81 patients, there were 114 bronchoscopies total. The mean number of DLBs per patient was 1.53 ± 0.50 . 42 patients had more than 1 DLB. The median time between tracheostomy and first DLB was 13 months (IQR 4.25-14.75). The median time between subsequent DLBs was 7 months (IQR 1.55-21.3). Ninety-five (83%) of these bronchoscopies were performed as outpatient procedures.

Sixty-six patients were diagnosed with airway findings on DLB (81.5%, Table 1). Among these suprastomal granulation was the most common finding (59.0%), followed by increased tracheal secretions (23%), stomal and peristomal granulation (13.2%), tracheal ulcer (3.3%), and suprastomal collapse (1.6%). The proportion of patients with airway findings who underwent DLB >6 months post-tracheostomy (90.6%) compared to <6 months post-tracheostomy (75.5%) was not significantly different ($p=0.087$). When examining only tracheostomy-related findings, the proportion of patients with airway findings who underwent DLB >6 months post-tracheostomy (61%) compared to <6 months post-tracheostomy (36%) was significantly different ($p = 0.026$).

Fifty-six (49.1%) DLBs were conducted due to patient symptoms. This equaled 37 patients who were symptomatic before surveillance DLB during the study period. The most common indications for interventional DLB were respiratory distress (45%), continued ventilator dependence (38%), and bleeding (18%). Fifty-eight DLBs were done when patients were not symptomatic and were thus purely surveillance. Patients who were symptomatic before DLB were significantly more likely to have positive findings on DLB (91.9%) as compared to patients who were asymptomatic (i.e., surveillance) prior to DLB (72.7%, $p=0.027^*$). Patients who were symptomatic before DLB were significantly more likely to have

tracheostomy-related airway findings on DLB (64.8%) as compared to patients who were asymptomatic (36.3%, $p = 0.011$).

Patients were significantly more likely to be symptomatic if they had DLB >6 months after tracheostomy versus <6 months after tracheostomy (68.8% vs. 30.6%, $p < 0.001^*$). Patients with symptoms before first DLB were more likely to require repeat bronchoscopy (95.8% vs. 75.4%, $p=0.003^*$), although the time interval between recurrent DLBs was not significantly different between patients who were symptomatic at the time of their first DLB and those who were asymptomatic.

There was no significant difference in the number of airway findings depending on the original indication for tracheostomy ($p=0.42$). Overall, airway findings were noted in 100% of patients with congenital upper airway obstruction, 67.7% of patients with TBM, 65.4% of ventilator dependent patients, and no patients with bronchopulmonary dysplasia. Of note, the airway findings noted in patients with congenital upper airway obstruction were new lesions and not those present at time of tracheostomy. When examining only tracheostomy-related findings, there was not a significant difference in the number of airway findings depending on the original indication for tracheostomy ($p = 0.85$).

Out of the 114 DLBs performed, 19 (16.6%) required operative interventions during the DLB. There were 32 total interventions, with 13 DLBs requiring more than one intervention. In other words, only 17 patients with findings noted on DLB requiring interventions at the time of the DLB (20.9). Out of the 37 patients who were symptomatic, 15 (40.5%) required intervention at the time of DLB. For tracheostomy-related complications, the most common interventions were removal of suprastomal granulation or dilation of subglottic stenosis (28.6%), removal of stomal granulation (21.4%), tracheostomy tube size change (14.3%), and removal of subglottic cyst (7.1%). For non-tracheostomy related complications, the most common interventions were tonsillectomy and/or adenoidectomy done for failed capped PSG (43.7%), tracheostomy tube size change (39.1%) and dilation of preexisting subglottic stenosis (17.4%).

The median number of months from tracheostomy procedure to decannulation was 36 months (IQR 29.0-62.5), and 23 of the 81 patients were decannulated at last follow-up. DLB was performed prior to decannulation and patients who had OSA on capped PSG also underwent adenotonsillectomy at the time of that DLB.

Finally, use of different tracheostomy tube types did not affect the number of airway findings. There was no significant difference between FlexTend tracheostomy versus standard tracheostomy ($p=0.76$) or cuffed versus uncuffed tubes ($p=0.23$). The number of airway findings did not significantly differ depending on age at which patients underwent tracheostomy ($p=0.30$), gender ($p=0.39$), term pregnancy ($p=0.69$), or continued ventilation support after tracheostomy ($p=0.54$).

4. Discussion

Despite evidence that the rate of post-tracheostomy complications can be as high as 63% in children, there are no current standardized clinical practice guidelines for the use of

bronchoscopy or laryngoscopy (DLB) in this population [14]. This study examined the utility of DLBs in children with tracheostomies by quantifying and comparing the incidence of findings and need for intervention. We observed a high rate of airway findings (Table 1), supporting the utility of DLB post-tracheostomy. Accordingly, patients were more likely to be symptomatic if they had a DLB >6 months after tracheostomy (Table 1). Symptomatic patients were more likely to have both overall airway findings (Table 1) as well as tracheostomy-related findings. When examining only tracheostomy-related findings, the proportion of patients with airway findings who underwent DLB >6 months post-tracheostomy compared to <6 months post-tracheostomy was significantly different. Finally, over 20% of patients required an intervention at the time of their DLB.

The most common indication for tracheostomy in our population was ventilator dependence for chronic lung disease or neuromuscular disorder, followed by airway obstruction due to either a congenital structural anomaly or TBM. These are consistently cited as the most common indications for pediatric tracheostomies, thus the findings from this study can be generalized to larger populations [4, 7, 15, 16]. We did not find a higher rate of overall airway findings or tracheostomy-related findings on DLB based on indication for tracheostomy (Table 1). Gergin et al. [6], on the other hand, found that patients with ventilator support and cardiopulmonary or traumatic indications had higher rates of lesions diagnosed on DLB. Our findings, however, indicate that the decision to pursue DLB should not be dictated by indication for tracheostomy and that ventilator dependent children are not necessarily at highest risk for positive findings on DLBs. Given this incongruity, further research is necessary to understand if the original indication for tracheostomy placement affects future DLB findings, which would make it important to potential guidelines for surveillance DLB.

In line with prior studies [6, 7], we observed a high rate of airway findings during DLB (Table 1). When further classifying complications as tracheostomy-related, there was still a high rate of airway findings with suprastomal granulation and increased tracheal secretions being the most common. This assures that the findings were not always previously identified and simply persistent, but, more likely a result of the tracheostomy. Our rate of suprastomal granulation was comparable to that reported in prior studies (59.0% compared to 39.9% to 44%) [6, 7]. In the future, it would be useful to relate this finding to tracheostomy type (i.e., FlexTend, Bivona, Shiley) to determine how this might impact granulation tissue formation. However, we did not find any difference amongst the various tracheostomy tube types. But, our smaller sample size may not have been sufficiently powered to find such a potential difference between tracheostomy types. Of note, all of our most common tracheostomy-related complications are considered “long-term complications.” These are consistently seen more often than short-term complications, even in prior work, highlighting the importance of interval follow-up after tracheostomy [1, 7, 16].

The most common indications for DLB were respiratory distress, continued ventilator dependence, and bleeding. Of note, in our population, nearly half (49.1%) of DLBs were preceded by symptoms, observed in clinic or in the hospital, prior to endoscopic evaluation. In prior work, a similar percentage (23% to 48.2%) of patients were symptomatic [6, 7].

This may be explained by the fact that many clinicians based the decision to pursue DLB on the presence of symptoms rather than on a routine basis.

Additionally, we found that patients who were symptomatic prior to DLB were more likely to have positive airway findings as compared to DLBs that were done for asymptomatic patients (i.e., surveillance). Previous studies have demonstrated mixed results with regards to the predictive value of symptoms prior to DLB [6, 7]. Intuitively, it makes sense that symptomatic patients are more likely to have lesions. This is an important relationship because it may guide a clinician's decision to pursue DLBs in patients who are symptomatic if evidence shows that symptoms increase the likelihood ratio of discovering airway findings. Additionally, symptomatic patients were more likely to have tracheostomy-related findings which helps parse out findings that may have already been present. On the other hand, symptomatic patients were *not* more likely to require interventions. The fact that identifying an airway finding might not necessarily lead to a change in clinical management lessens the potential value of this procedure. Indeed, operative intervention may actually lead to worsening symptoms and complications such as bleeding, inflammation, and heightened risk of infection.

Of patients who underwent DLB, 20.9% required an intervention during endoscopic evaluation. The rate of intervention in prior studies ranged from 33.3% to 56.6% [6, 7]. While the majority of our patients did not require an intervention at the time of DLB, endoscopic findings may have spurred closer monitoring and non-operative management. Examples include medication management for reflux or the use of nebulized steroids for granulation tissue and inflammation. The decision to pursue intervention at the time of DLB is controversial and recent work has suggested that it should be made on a case-by-case basis [4, 7]. For tracheostomy-related complications, the most common interventions were removal of suprastomal granulation, dilation of subglottic stenosis, removal of stomal granulation, and tracheostomy tube size changes. For non-tracheostomy related complications adenoidectomy and/or tonsillectomy (TA) was the most common intervention which was independent of tracheostomy status. The majority of these are done for patients who failed an initial capped tracheostomy sleep study in preparation for decannulation. In these cases, the TA was done to reduce airway obstruction. The fact that need for intervention did not differ between symptomatic and asymptomatic patients suggests that the presence of symptoms prior to DLB may not predict subsequent need for operative intervention. This also indicates that, in some cases, conditions such as suprastomal granulation tissue, subglottic stenosis or tracheostomy size mismatch may be asymptomatic. In these cases, managing complications prior to symptom presentation may prevent later tracheostomy-related complications.

Prior work did not find a significant difference in the incidence of airway findings depending on timing of DLB post-tracheostomy [6, 7]. We noted more overall lesions in patients who underwent endoscopy >6 months post-tracheostomy compared to those who underwent endoscopy <6 months post-tracheostomy, although this did not reach significance (Table 1). Interestingly when we examined only tracheostomy-related findings, the proportion of patients with airway findings who underwent DLB >6 months post-tracheostomy was significantly higher than those who underwent DLB <6 months post-tracheostomy. This

suggests that a longer interval after tracheostomy placement may be necessary to appreciate airway lesions that are incited by tracheostomy placement. In addition, continuing interval DLB after 6 months may allow us to capture findings related to the tracheostomy.

Patients were significantly more likely to be symptomatic if they had DLB >6 months after tracheostomy versus <6 months after tracheostomy (Table 1). This result may be attributed to the fact that the majority of our airway findings were considered “late” complications (e.g., suprastomal granulation, increased tracheal secretions) and therefore may take time to develop and become subsequently symptomatic. For example, patients may develop more suprastomal granulation tissue over time which eventually leads to obstruction. As such, they may not be immediately aggravated by tracheostomy placement. Taken together with the fact that tracheostomy-related airway findings were more common within 6 months indicates that waiting longer than this to pursue DLB might be too long.

Our study examined the effect of tracheostomy type on the overall incidence of tracheostomy related airway findings, which has not been studied previously. We did not find a difference in lesions across tracheostomy type. However, given that only 9.9% of our tracheostomy tubes were Flextend type tubes, these findings may not be representative and larger sample sizes are needed. Similarly, the rate of airway findings did not differ by sex, prematurity or age. This has been documented in prior studies and indicates that these specific patient characteristics may not be as useful when considering what patients would benefit most from DLB [6, 7].

Another factor that might impact the incidence of tracheostomy-related complications such as infection or granulation is the tracheostomy care routine. At our institution, all patients/families undergoing tracheostomy are taught a standard protocol of tracheostomy care which includes the following instructions: (1) Change the tracheostomy tube on a weekly basis unless the patient is experiencing increased secretions or mucous plugging, in which case the tube may be changed sooner (2) Receive one new tracheostomy a month per Medicaid (3) Discard tracheostomies after three months (4) Clean tracheostomy per manufacturer guidelines. There are no recommendations provided regarding specific dressing changes for the flanges.

Additionally, all patients are required to demonstrate competency with tracheostomy changes and cleaning prior to their discharge from the hospital. Since this is a standardized routine and licensed respiratory therapists confirm competency in all patients, we do not believe that a comparison in care routines is necessary between groups (e.g., those with airway findings and those without).

Our study has several limitations. While this is a single institution study, the range of practice patterns among our pediatric otolaryngologists mimics the diversity of practice across the field. Finally, our sample size is limited compared to the two previously published studies [6, 7]. However, the existing data is also limited, and any additional perspective will bolster an understanding of how DLBs may best fit into clinical care.

5. Conclusions

The results of this study answer a call to action for further research examining the utility of DLBs in pediatric tracheostomy [3, 7]. Given the high incidence of airway findings noted on DLB, we corroborate previous claims that endoscopic evaluation is useful in pediatric patients with tracheostomies. Specifically, DLB is most valuable in symptomatic patients and clinicians should have a low threshold in pursuing DLBs in this cohort. Additionally, scheduled DLB is a vital component of long-term interval follow-up in children with tracheostomies, as evidenced by the higher rates of complications in patients >6 months post-tracheostomy. To summarize, symptomatology and post-tracheostomy timing must guide the use of DLB in pediatric patients with tracheostomies. Adherence to the above recommendations provides clear circumstances in which DLBs are appropriate and reinforces the movement away from unnecessary anesthetic exposure in developing pediatric patients.

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Table 1.

Airway findings observed during DLB based on patient and clinical characteristics.

		Airway Lesion Present (n/total)	%	P-value
All		66/81	81.5%	
Time to DLB				0.087
	<6 months	37/49	75.5%	
	>6 months	29/32	90.6%	
Symptoms before DLB				0.027*
	Yes	34/37	91.9%	
	No	32/44	72.7%	
Indication for tracheostomy				0.42
	Ventilator dependence	34/52	65.4%	
	Tracheobronchomalacia	20/30	66.7%	
	Respiratory failure	13/14	92.9%	
	Vocal cord paralysis	4/5	80.0%	
	Congenital upper airway obstruction	3/3	100.0%	
	Sleep apnea	2/2	100.0%	
	Bronchopulmonary dysplasia	0/1	0.0%	
Age at tracheostomy				0.30
	0-5 months	23/39	59.0%	
	6-11 months	14/16	87.5%	
	1-9 years	17/25	68.0%	
	10-18 years	1/1	100.0%	
Continued ventilation support after tracheostomy				0.54
	Yes	23/27	85.2%	
	No	43/54	79.6%	
Sex				0.39
	Female	30/35	85.7%	
	Male	36/46	78.3%	
Preterm				0.69
	Term	37/43	91.9%	
	Preterm (<40 weeks)	13/16	81.3%	

DLB=Diagnostic laryngoscopy and bronchoscopy