# Impact of Tracheobronchopathia Osteochondroplastica on Difficult Intubation

# Mattea E. Miller, $BS^{1}$ , Ioan Lina, $MD^{1}$ , Sara Sateri, $MD^{2}$ , and Nasir Bhatti, MBBS, $MD^{1}$

#### Keywords

airway management, difficult intubation, erosive nasal disease, tracheobronchopathia osteochondroplastica

Received August 14, 2023; accepted August 18, 2023.

racheobronchopathia osteochondroplastica (TPO) is a rare condition of the trachea and bronchus characterized by the development of submucosal osteocartilaginous nodules that spare the posterior tracheal wall, a characteristic used for diagnosis of TPO.<sup>1,2</sup> Symptoms of TPO are nonspecific and most commonly include chronic cough, wheezing, and dyspnea on exertion although clinical manifestations are highly variable and can lead to unanticipated difficult intubation in surgical settings.<sup>3</sup> Recognizing patients at risk of TPO is of utmost importance as failure to obtain a secure airway can result in significant complications.<sup>4</sup> Here, we present a case in which TPO was diagnosed after the inability to intubate during elective nasal endoscopy in the operating room. This study was approved by the Johns Hopkins Institutional Review Board (IRB00366662).

# **Case Report**

A 43-year-old woman with a history of asthma, allergies, and chronic dyspnea on exertion presented with a 15-year history of sinus pain, ear pain, headaches, odynophagia, nasal congestion, and postnasal drip. She denied any previous nasal surgery or trauma. Her initial office visit was remarkable for sinus congestion, nasal drainage nosebleeds, and crusting in both nostrils. In-office nasal endoscopy revealed severe crusting along the inferior and middle turbinates bilaterally and associated erosion of the lateral nasal walls. Computed tomography (CT) sinus was notable for erosive/destructive changes of the ethmoid sinuses, middle and inferior turbinates bilaterally, and moderate mucosal thickening throughout the paranasal sinuses. After failure of maximal medical therapy, she was scheduled for endoscopic debridement and biopsy for possible chronic inflammatory/destructive pathology.



F O U N D A T I O N OTO Open 2023, Vol. 7(3):e81 © 2023 The Authors. OTO Open published by Wiley Periodicals LLC on behalf of American Academy of Otolaryngology-Head and Neck Surgery Foundation

DOI: 10.1002/oto2.81 http://oto-open.org WILEY

After induction of anesthesia, direct laryngoscopy was performed sequentially by 2 experienced operators who obtained direct visualization of the glottic opening; however, were unable to pass a 65 mm endotracheal tube (ETT) past the glottis due to subglottic resistance. Under indirect visualization with a video laryngoscope, the distal tip of the ETT was placed just at the glottic opening and the anesthesia team was able to ventilate the patient despite suboptimal and unsecured ETT placement. A flexible bronchoscope was passed through the ETT to visualize the subglottic obstruction. Resistance was encountered and the tracheal lumen was not able to be effectively visualized. The surgery was aborted, and the patient was awoken from surgery without complications.

A week later flexible laryngoscopy in the clinic confirmed significant granulation tissue in the subglottic area with intact vocal cord motion; however, the lower trachea could not be visualized. CT chest revealed nodular irregularity and calcifications of trachea sparing posterior wall, with associated luminal narrowing, most notable at the subglottic airway (transverse diameter 3 mm), and otherwise normal caliber bronchi (**Figure 1**). She was placed on a prednisone taper without substantial clinical improvement in her symptoms. Additional workup included erythrocyte sedimentation rate, C-reactive protein, and urinalysis all within normal limits, antineutrophil cytoplasmic autoantibody, cytoplasmic (c-ANCA) 1:320; and negative anti-MPO and anti-PR3 antibodies.

Given suspicion for TPO a bronchoscopy with interventional pulmonology was performed and revealed dense endobronchial disease with mildmoderate obstruction throughout the trachea and into the bilateral mainstems with sparing of the posterior

#### **Corresponding Author:**

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

<sup>&</sup>lt;sup>1</sup>Department of Otolaryngology-Head and Neck Surgery, Johns Hopkins University School of Medicine, Baltimore, Maryland, USA

<sup>&</sup>lt;sup>2</sup>Department of Anesthesiology and Critical Care Medicine, Johns Hopkins University School of Medicine, Baltimore, Maryland, USA

Nasir Bhatti, MBBS, MD, Johns Hopkins University School of Medicine, 1830 East Monument Street, Suite 7300, Baltimore, MD 21205, USA. Email: nbahatti@jhmi.edu



Figure 1. (A) Endoscopic view of the subglottic trachea, (B) CT without contrast sagittal rendering, (C) CT without contrast coronal rendering, (D) CT without contrast sagittal, (E) CT without contrast axial, and (F) CT without contrast axial. CT, computed tomography.



Figure 2. Histologic slides from an endobronchial biopsy of the right mainstem bronchus showing squamous and respiratory mucosa with underlying bone, acute and chronic inflammation, and keratinous debris. Grocott methenamine silver stain was negative for fungus.

wall (Figure 1). Endobronchial biopsies were taken from the right mainstem which revealed squamous and respiratory mucosa with underlying bone, acute and chronic inflammation, and keratinous debris (Figure 2). Grocott methenamine silver stain was negative for fungus.

# Discussion

Herein we report the first case reported in the literature of TPO diagnosed secondary to a failure of intubation during intraoperative assessment of a destructive nasal pathology.

Diagnosis of TPO can be made by CT or bronchoscopy, yet is often incidentally found during intubation, or during autopsy.<sup>2,3</sup> CT findings characteristic of TPO include nodular submucosal calcifications sparing the posterior tracheal wall.<sup>2,3,5</sup> In our patient, concurrent nasal pathology with elevated c-ANCA titers suggests that additional manifestations of TPO may exist and is an important consideration during surgical planning to avoid morbidity and mortality.<sup>4</sup> In non-emergent cases during elective procedures, maintenance of spontaneous ventilation and patient emergence from anesthesia should be a priority when considering possible TPO.

# Conclusion

TPO is a rare benign condition that results from the development of osteochondral nodules and can cause acute airway compromise and unanticipated difficult intubation in patients presenting with otolaryngologic complaints.

# **Author Contributions**

Mattea E. Miller, conducted data collection, interpreted the data, drafted the initial manuscript, and reviewed the manuscript; Ioan Lina, conceptualized the study, interpreted the data, and reviewed the manuscript; Sara Sateri, reviewed the manuscript; Nasir Bhatti, conceptualized the study, interpreted the data and reviewed the manuscript. All authors approved the final manuscript as submitted and agreed to be accountable for all aspects of the work.

# Disclosures

**Competing interests:** The authors report no conflicts of interest. **Funding source:** None.

# ORCID iD

Mattea E. Miller D http://orcid.org/0000-0001-5649-9996 Ioan Lina D http://orcid.org/0000-0002-8619-7312 Nasir Bhatti D http://orcid.org/0000-0002-3355-0549

#### References

- Fois AG, Arcadu A, Santoru L, et al. Tracheobronchopathia osteochondroplastica: a rare case report of a non-smoker and non-atopic patient, with a long history of wheezing since childhood. *Multidiscip Respir Med.* 2016;11:16. doi:10.1186/ s40248-016-0050-7
- Jabbardarjani HR, Radpey B, Kharabian S, Masjedi MR. Tracheobronchopathia osteochondroplastica: presentation of ten cases and review of the literature. *Lung.* 2008;186(5): 293-297. doi:10.1007/s00408-008-9088-4
- Wang W, Hu H, Liu M, Wang J. Tracheobronchopathia osteochondroplastica: five cases report and literature review. *Ear Nose Throat J.* 2020;99(10):NP111-NP118. doi:10.1177/ 0145561319897982
- 4. Kar S, Senapati LK, Samanta P, Satapathy GC. Predictive value of modified Mallampati test and upper lip bite test concerning Cormack and Lehane's laryngoscopy grading in the anticipation of difficult intubation: a cross-sectional study at a Tertiary Care Hospital, Bhubaneswar, India. *Cureus*. 2022;14(9):e28754. doi:10.7759/cureus.28754
- 5. Sharma D, Kamath S, Acharya V, Rai S. Pebbled trachea: a case series and literature review of tracheobronchopathia osteochondroplastica. *J Glob Infect Dis.* 2021;13(4):180. doi:10.4103/jgid\_jgid\_379\_20