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Abstract

Introduction

Airway enlargement was first described histologically in 1872, but it was not clinically reported until 1932, when Dr. Mounier-Kuhn published his seminal radiographic studies.¹ Since then, it is estimated that several hundred case reports have been published on congenital Mounier-Kuhn Syndrome (MKS), acquired tracheomegaly, and tracheobronchomegaly (TBM), yet only a few comprehensive literature reviews exist.²

Case

This case report details an elective nasal procedure for a patient with a history of significant pulmonary hypertension and COPD on 5L home O₂ who had a symptomatic facial trauma. He was considered high risk secondary to his pulmonary disease, but he was considered medically optimized for surgery and had undergone a prior uncomplicated general anesthetic. An intraoperative endotracheal tube dislodgement necessitated emergency airway rescue with difficulty in re-establishing adequate ventilation resulting in the patient being brought to the ICU post-operatively. After a second endotracheal dislodgement shortly later, retrospective radiographic airway analysis revealed tracheal dimensions of 32 x 26mm, suggesting a previously undiagnosed tracheomegaly. Written consent and HIPAA authorization was obtained from the patient for the publication of this case report.

Conclusion

This case highlights the clinical importance for anesthesiologists to be aware of tracheobronchomegaly (TBM) despite limited provider experience with these patients and sparse reporting on the condition. It is underdiagnosed radiographically and may first present in the OR. TBM can manifest in many ways with potentially fatal complications thus it is vital for anesthesiologists to be able to recognize, troubleshoot, and potentially treat issues related to the disease.

Keywords

Tracheobronchomegaly, tracheomegaly, Mounier-Kuhn Syndrome, endotracheal dislodgement, extubation, air leak, pressure leak, airway emergency, difficult airway

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Endotracheal Tube Dislodgment in a High-Risk Pulmonary Hypertensive Patient due to Undiagnosed Tracheomegaly: A Case Report

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This case highlights the clinical importance for anesthesiologists to be aware of tracheobronchomegaly (TBM) despite limited provider experience with these patients and sparse reporting on the condition. It is underdiagnosed radiographically and may first present in the OR. TBM can manifest in many ways with potentially fatal complications thus it is vital for anesthesiologists to be able to recognize, troubleshoot, and potentially treat issues related to the disease.

Introduction

Airway enlargement was first described histologically in 1872, but it was not clinically reported until 1932, when Dr. Mounier-Kuhn published his seminal radiographic studies.¹ Since then, it is estimated that several hundred case reports have been published on congenital Mounier-Kuhn Syndrome (MKS), acquired tracheomegaly, and

tracheobronchomegaly (TBM), yet only a few comprehensive literature reviews exist.²

This case presents one of the first known reports of endotracheal tube dislodgement from undiagnosed TBM, resulting in airway emergency, during a nasal procedure in a high-risk pulmonary hypertensive patient. It is intended that this case highlight the serious potential complications of TBM, to increase the anesthesiologist's awareness and understanding of this highly relevant and underreported disease.

Case

A 62-year-old male (height 1.85 m, weight 86.5 kg) with group 1 pulmonary artery hypertension presented for rhinoplasty and septoplasty due to facial trauma, nasal obstruction, septal deviation, and nasal valve collapse. His medical history was significant for oxygen dependent COPD on 5L home O₂ with arterial oxygen saturations (SpO₂) ranging from 85-93%, decreased exercise tolerance, acquired polycythemia, depression, and spinal stenosis with chronic back pain. His anesthetic history was unremarkable, with a previous uncomplicated procedure under general anesthetic five years ago.

The patient was deemed high risk given his significant pulmonary disease and was therefore evaluated pre-operatively by a pulmonologist who felt that the patient was optimized for surgery. Despite the increased risk, the patient elected to proceed with surgery given his decreased quality of life related to his nasal trauma. Physical examination revealed a Mallampati III airway with unremarkable cardiac and pulmonary exams. A perioperative transthoracic echocardiogram revealed a moderately dilated RV with normal function, however there was insufficient TR to estimate pulmonary artery systolic pressure (PASP). Right heart catheterization was requested but deferred.

Following pre-oxygenation and placement of standard ASA monitors, general anesthesia was induced with fentanyl 50 mcg IV, propofol 160 mg IV, and rocuronium 50 mg IV, and the patient was atraumatically intubated with an 8.0 mm endotracheal tube (ETT). General anesthesia was maintained with sevoflurane at 0.7 MAC, and ventilator

settings were adjusted to maintain SpO₂ > 85% and EtCO₂ was maintained between 32 – 35 mmHg.

The case proceeded normally, however after roughly 90 minutes; the patient's tidal volume abruptly dropped to approximately 20 mL with loss of the EtCO₂ reading. The patient could not be manually ventilated, and it was suspected that the ETT had migrated. Direct laryngoscopy revealed what appeared to be a properly placed ETT with the cuff below the cords, however, with arterial saturations dropping, the decision was made to remove the existing ETT and replace it with a new 7.5 mm ETT. Of note, the initial endotracheal tube was found to be in good condition with no damage to the cuff. EtCO₂ tracing returned, however, there was a persistent cuff leak with the new ETT. Video laryngoscopy revealed proper positioning of the ETT with the cuff below the cords. Additional air was added to the cuff (total of 15 mL) with resolution of the leak. Ventilation recovered though arterial saturation remained below baseline. Bronchoscopy was performed which revealed a small amount of blood in the proximal airway. Epoprostenol 0.16 mg/hr was begun with improvement of oxygen saturation to baseline. At the completion of the procedure, the patient was taken to the ICU intubated and sedated on propofol infusion to continue the epoprostenol.

Approximately eight hours after arriving in the ICU, the patient developed a new large cuff leak with all respiration occurring spontaneously around the ETT. Tube depth and positioning was confirmed with X-ray and by visual inspection. The addition of air to the cuff and attempts to advance the tube, did not resolve the large leak, so the patient was extubated to face mask with stable SpO₂. Over the next two days he was transferred out of the ICU, weaned to his baseline oxygen requirement, and discharged home in good condition.

Due to the multiple dislodgements of the patient's ETT throughout this hospitalization, his records were reviewed more thoroughly by the anesthesia team. A CT of the chest from four years prior to this admission was discovered that revealed tracheal dimensions of 32 x 26mm. This had not been mentioned in the CT report, and the patient was subsequently diagnosed with tracheomegaly.

Discussion

Mounier-Kuhn Syndrome or congenital tracheomegaly/tracheobronchomegaly (TBM) is a rare condition with an unknown prevalence. However, there are an estimated 200-300 case reports in the literature. TBM is typically diagnosed after the third decade of life, with various reports citing a 4:1 – 8:1 male predominance.^{2,3} Though the exact mechanism is unknown, it is believed to originate from a loss of smooth muscle and elastic fibers in the conducting airways, causing diverticular projections, bronchiectasis, impaired mucociliary clearance, and overly compliant walls which are prone to collapse.⁴⁻⁷

Patients may present with nonspecific cough, frequent pulmonary infections, dyspnea, hemoptysis, clubbing, respiratory insufficiency, obstruction, or cor pulmonale.^{2,4,8} Differential diagnoses and secondary causes include connective tissue disorders, cutis laxa, pulmonary fibrosis, cystic fibrosis, emphysema, pneumonia, high cuff pressures, prolonged mechanical ventilation, barotrauma, ankylosing spondylitis, and light chain deposition disease.^{7,9,10} Treatments for TBM include medical management, such as bronchodilators and corticosteroids for symptom control as well as vaccinations for prophylaxis, and surgical options, such as stenting, tracheobronchoplasty, laser treatment, or lung transplant. As a man, in his 7th decade, on corticosteroids, with severe oxygen-dependent obstructive lung disease and pulmonary hypertension, our patient presented with typical TBM symptoms.

TBM can be diagnosed with X-ray or CT, but unfortunately, these measurements are not taken routinely.² Diagnostic TBM criteria for X-ray include a tracheal diameter > 3.0 cm measured 2 cm above the aortic arch, a right bronchus diameter >2.4 cm, and a left bronchus diameter >2.3 cm. On CT, which is the gold standard, men are diagnosed with transverse and sagittal tracheal measurements of >25 mm and >27 mm, respectively (women >21 mm and 23 mm), right bronchus diameter >21.1 mm, and left bronchus diameter >18.4 mm.^{4, 7, 11} Our patient had X-rays taken after his procedure, and his previous X-rays from 4 years prior were obtained for comparison. He had also had a CT performed 3 years prior to this surgery, with a representative cut shown in Figure 1 that reveals tracheal dimensions of approximately 32 x 26 mm. His bronchi did not meet criteria for enlargement, so his imaging was consistent with a diagnosis of tracheomegaly, but not tracheobronchomegaly

or Mounier-Kuhn. Had this diagnosis been made prior to his surgery, repeat imaging may have been useful for planning a more secured airway that would not be so easily lost, with plans for alternate intubation strategies.

Patient's with TBM pose unique anesthetic risks due to their large, weakened airways. Anesthetic complications that have been reported include severe cuff leak, endotracheal tube dislodgement, aspiration pneumonia, impaired cough, decreased secretion clearance, tracheal injury, tracheal diverticula, and airway dilation with inspiration/collapse with expiration.^{2,4,6,8,11} Strategies that can be employed in patients with known or suspected TBM include: using moist oropharyngeal packing to decrease air leaks and aspiration risk, careful suctioning, utilizing positive pressure to minimize air leaks, upsizing the ETT, considering LMAs as an alternative to ETT, and lung protective ventilation to prevent barotrauma.^{5,11,12} Although overinflating the ETT cuff may be a considered during a cuff leak, it is recommended to keep cuff volumes less than 6 – 8 cc's, as >10 cc raises concerns for tracheal injury.¹⁰ Repositioning the ETT to a narrower portion of the airway is also possible, especially when utilizing 3D tracheal reconstruction imaging and careful fiberoptic instrumentation. Given the variety of anesthetic risks and possible presentations, each situation will likely require its own unique considerations, but with a differential that included TBM and a basic understanding of the disease, these issues can be easily managed.

Conclusion

As this case highlights, TBM is highly relevant to anesthesiologists. TBM is underdiagnosed radiographically, and many patients may first present in the OR. From obstruction to air leak to endotracheal dislodgment, the disease can manifest in a multitude of ways and result in emergent or event fatal consequences. Vigilance and a high index of suspicion in at risk patients is imperative to ensure a good outcome. ■

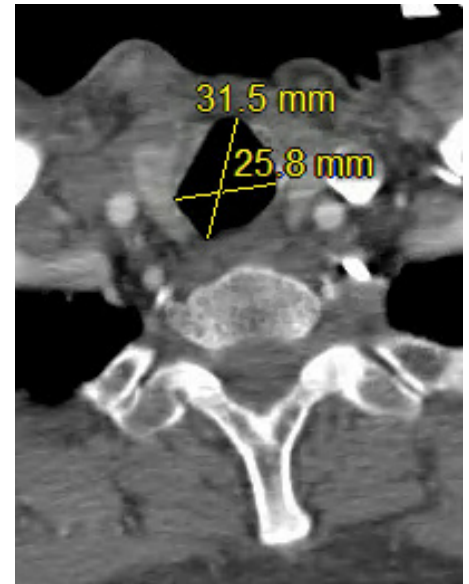


Figure 1. Representative image from patient's prior CT. The trachea measures 31.5 x 25.8 mm which is consistent with the diagnostic criteria for tracheomegaly. Measurements were not made until after the airway issues encountered in this case and were not included in the radiology report.

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