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Symptomatic Improvement in Cicatricial Pemphigoid of the Trachea Achieved with Laser Ablation Bronchoscopy

Elizabeth Benge, MD¹, Vincent Tran MD², Nazanin Sheikhan MD¹, Sapna Bhatia MD³, Yi McWhorter DO⁴, John Collier MD³, Arnold Chung MD⁵

Departments of ¹Internal Medicine, ²Surgery, ³Pulmonology, ⁴Anesthesiology/Critical Care Medicine, and ⁵MountainView Cardiovascular and Thoracic Surgery Associates
HCA Healthcare MountainView Hospital
Las Vegas, NV, USA

Abstract

Cicatricial pemphigoid (CP) with tracheal involvement is a rare and potentially deadly condition. Here, we report the first case in which Nd:YAG laser (1064nm) laser ablation bronchoscopy was used to treat CP with tracheal involvement. Our patient is a 71-year-old male with a history of CP refractory to medical therapy affecting his trachea who presented to the emergency department with dyspnea. He ultimately underwent bronchoscopy with Nd: YAG laser (1064nm) laser ablation, which resulted in a temporary alleviation of his respiratory symptoms. A repeat laser ablation was planned in hopes of prolonging the patient's remission, but due to interval changes in the patient's airway anatomy, it was deemed unsafe. While our patient's uniquely advanced disease was not amenable to further laser-mediated intervention, it is possible that patients with less advanced disease may experience better outcomes with similar therapy. This case shows the promise laser ablation could hold for patients with tracheal cicatricial pemphigoid.

Introduction

Cicatricial pemphigoid (CP) is a diverse group of subepithelial blistering disorders of the skin and mucous membranes (1,2). Tracheal involvement is a rare and deadly sequela of this disease class (3). We report the first case in which Nd:YAG laser (1064nm) laser ablation bronchoscopy was used as a treatment for CP with tracheal involvement. Of note, the terms cicatricial pemphigoid and mucous membrane pemphigoid are synonymous and

are used interchangeably throughout this report.

Case Presentation

Our patient is a 71-year-old man with a history of CP affecting his left eye and trachea who presented to the emergency department with progressively worsening dyspnea. The patient has a history of multiple bronchoscopies; the most recent one showed tracheal pemphigoid lesions partially obstructing his airway. His diagnosis of cicatricial pemphigoid had been made over

fifteen years prior to the current presentation via biopsy and subsequent immunofluorescence staining. On admission, his respiratory rate was 21 breaths/min and his oxygen saturation was 97% on 50% Bipap: 14/8. He was admitted to the intensive care unit for evaluation and management of his acute hypoxic respiratory failure.

Initially, a fiberoptic bronchoscopy was performed under laryngeal mask airway (LMA) general anesthesia. Dense, dark-colored lesions were noted to be occluding most of the trachea, consistent with the patient's history of tracheal CP (Figure 1).



Figure 1. Patient's trachea demonstrating heavy burden of cicatricial pemphigoid lesions prior to any intervention

They were partially removed in a piecemeal manner with forceps instrumentation. After this procedure, the patient still required supplemental oxygen, oscillating between BiPAP and nasal cannula. Two days later, he was started on rituximab, which he had also received during previous relapses.

On hospital day four, our cardiothoracic surgery team performed bronchoscopy with laser ablation under LMA general anesthesia. After the procedure, the patient's tracheal

lesions had markedly decreased in size (Figure 2).



Figure 2. Patient's trachea with reduced lesions status-post bronchoscopy with laser ablation.

He was also entirely weaned off supplemental oxygen.

In the following weeks, the patient's symptom burden was significantly decreased. He reported an improvement in his quality of life and satisfaction with the procedure. A subsequent repeat laser ablation was planned at the three-month mark. This procedure was more technically challenging due to airway-narrowing caused by an increase in scar tissue from the initial laser ablation. Due to the risks imposed by the interval changes in the patient's anatomy, we decided against further laser therapy. In the absence of laser treatments, the patient's tracheal pemphigoid recurred and symptoms returned to their prior state. He currently receives interval fiberoptic bronchoscopies to partially remove his lesions when they threaten his airway.

Discussion

In a study involving subjects with aggressive ocular CP, 81% of patients achieved clinical remission with rituximab therapy (4). Medical therapy had repeatedly failed to reduce our

patient's symptoms, making his case unique in both its rarity and refractory nature. With no other options, our team developed an innovative treatment modality in an attempt to offer our patient some symptomatic relief.

Previous case reports have shown the utility of low-level laser therapy in mucous membranous lesions (5-7). One study showed successful resection of an obstructive mass caused by CP and restoration of airway patency using a Holmium LASER (2100nm) (8-9). We decided to ablate/resect the inflammatory tissue using an Nd:YAG LASER (1064nm) given its medium penetration length (1-4mm), coagulopathic ability (high heme absorption), and decreased tissue destruction when compared to the Ho:YAG laser; which has a higher laser absorption coefficient with water.

To our knowledge, this is the first case report of successful treatment of cicatricial pemphigoid with Nd:YAG laser (1064nm) ablation therapy. This procedure resulted in immediate, although ultimately impermanent, improvement in our patient's respiratory insufficiency. Our patient also reported an improved quality of life during the period of time the laser ablation therapy offered him symptomatic relief. He was able to attend his grandchildren's soccer games and walk to the end of his driveway to get his newspaper, activities he had not be able to participate in for years.

While our patient's improvement was temporary, his disease process was uniquely advanced. It is possible that patients with less advanced disease may experience longer periods of remission with laser-mediated therapy, or may be able to tolerate repeated laser ablation procedures. Importantly, our patient's case demonstrates that laser therapy can significantly reduce the burden of pemphigoid lesions, and can lead to a better

quality of life for a disease process with few alternative treatment modalities.

Conclusion

Therapeutic fiberoptic bronchoscopy with laser ablation is a promising treatment for patients suffering from CP of the trachea. Future investigations should focus on optimizing the laser ablation technique to achieve safe and sustained results.

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Abbreviations

- Bipap: bilevel positive airway pressure
- CP: cicatricial pemphigoid

- Ho:YAG: holmium-doped yttrium aluminum garnet
- Laser: light amplification by stimulated emission of radiation
- LMA: laryngeal mask airway
- Nd:YAG: neodymium-doped yttrium aluminum garnet

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