

Case Report

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

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Received: November 05, 2021; Accepted: November 30, 2021; Published: December 07, 2021

Abstract

Cicatricial Pemphigoid (CP) are a diverse group of subepithelial blistering disorders of the skin and mucous membranes. 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 with dyspnea. He initially underwent a fiberoptic bronchoscopy with piecemeal resection of his tracheal lesions, which mildly improved his symptoms. Next, he underwent bronchoscopy with Nd:YAG laser (1064nm) laser ablation. He was then entirely weaned off supplemental oxygen. For the following three months, he experienced a decreased symptomatic burden. A repeat laser ablation was planned, but due to interval changes in the patient's airway anatomy, it was deemed unsafe. Consequently, the patient's tracheal pemphigoid fully recurred. This case demonstrates the promise laser ablation holds for patients with tracheal cicatricial pemphigoid.

Keywords: Case report; Cicatricial pemphigoid; Laser ablation bronchoscopy

Abbreviations

CP: Cicatricial Pemphigoid; LMA: Laryngeal Mask Airway

Introduction

Cicatricial Pemphigoid (CP) are a diverse group of subepithelial blistering disorders of the skin and mucous membranes [1]. Tracheal involvement is a rare and deadly sequelae of this disease class [2]. We report the first case in which Nd:YAG laser (1064nm) laser ablation bronchoscopy was used as a treatment for CP with tracheal involvement.

Case Presentation

Our patient is a 71-year-old male 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. 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). 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.



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

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). 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

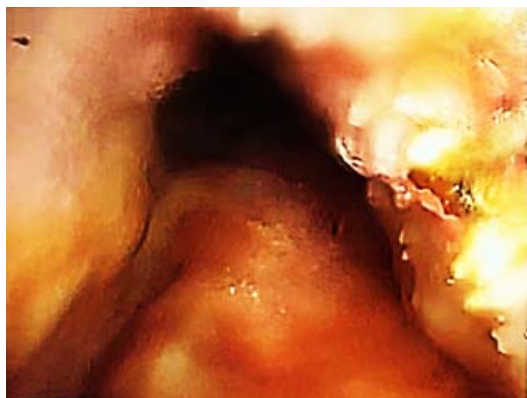


Figure 2: Patient's trachea with reduced lesions status-post bronchoscopy with laser ablation.

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 [3]. 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 [4-6]. One study showed successful resection of an obstructive mass caused by CP and restoration of airway patency using a Holmium LASER (2100nm) [7]. 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. While our patient's improvement was temporary, this case shows the promise laser ablation holds for other patients with tracheal cicatricial pemphigoid. We demonstrated that laser therapy can 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.

Funding

This research was supported (in whole or in part) by HCA Healthcare and/or an HCA Healthcare affiliated entity. The views expressed in this publication represent those of the author(s) and do not necessarily represent the official views of HCA Healthcare or any of its affiliated entities.

This case was presented at the CHEST Annual Meeting that took place from Oct 17, 2021 - Oct 20, 2021 in a virtual format.

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