

## Case Report

### Endobronchial leiomyoma successfully treated with flexible bronchoscopic cryotherapy: A case report

<sup>1</sup>Nepal SK, <sup>1</sup>Cirino M, <sup>2</sup>Grider D, <sup>1</sup>Rubio E

<sup>1</sup>Department of Pulmonary and Critical Care Medicine, Virginia Tech Carilion School of Medicine,

<sup>2</sup>Department of Pathology, Virginia Tech Carilion School of Medicine

#### Abstract

Benign tumors of the tracheobronchial tree are rare. Leiomyoma is one such tumor that, though benign, may lead to complete bronchial obstruction. Secondary symptoms are dependent upon the location and extent of airway involvement. The mainstay to manage these tumors has been surgical resection. With the advance in interventional bronchoscopic techniques, non-surgical options are being more commonly explored, including a few reports of successful resection of leiomyomas with laser therapy. We report the first case of complete resection of bronchial leiomyoma with endobronchial cryotherapy.

*Keywords:* Endobronchial leiomyoma, Cryotherapy, Flexible bronchoscopy

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#### Introduction

Endobronchial leiomyomas are rare benign tumors compromising about 2% of the excised benign endobronchial lesions. These tumors can lead to complete bronchial obstruction and secondary symptoms are dependent upon the location and extent of airway involvement. The mainstay to manage these tumors has been surgical resection. Non-surgical options are being more commonly explored, including a few reports of successful resection of leiomyomas with laser therapy. Although Cryotherapy is safe and effective for selective endobronchial tumors including endobronchial leiomyoma, this is underutilized.<sup>1,2</sup>

#### Case Report

A 53 year old, non-smoker, white woman presented with episodic wheezing and recurrent pneumonias (7 episodes between 2005 and 2014). She reported progressive shortness of breath on exertion, developing

non-specific intermittent right chest pain for past 2 months. She reportedly had asthma since childhood, treated with as needed albuterol.

Computed chest tomography (CT) (Figure 1) showed a lesion obstructing the bronchus intermedius. Flexible bronchoscopy confirmed almost complete bronchus intermedius obstruction by a white, smooth tumor with a broad base (Figure 2). A flexible 2.4 mm cryoprobe (ERBE, Marietta, Georgia, USA) with nitric oxide as a cryoagent, was subsequently used to remove the tumor. The probe was attached to middle of the lesion (to avoid damage to the surrounding airway mucosa) and then pushed towards its center. The probe was then frozen until there was a rim of icing, approximately 2-3 mm around the probe. Then, the bronchoscope was removed en-block with the tumor attached to the probe. The tumor measured 1 cm and felt hard. The tumor base had mild residual abnormal mucosa (Figure 2)

**Corresponding Author:** Nepal SK, Department of Pulmonary and Critical Care Medicine, Virginia Tech Carilion School of Medicine. Email: santoshnpl@gmail.com

measuring about 1 cm in the anterolateral wall of the bronchus intermedius. This was treated with 11 freeze-thaw cycles of 60 second each. Blood loss was <2 ml.

Histopathology (Figure 3) showed endobronchial mucosa with an underlying submucosal spindle cell proliferation with cigar shaped nuclei and perinuclear vacuoles in fascicular pattern, positive on a muscle specific action immunohistochemically stained tissue section, confirming the diagnosis of leiomyoma. No significant cellular atypia noted and no mitotic figures were identified, excluding leiomyosarcoma.

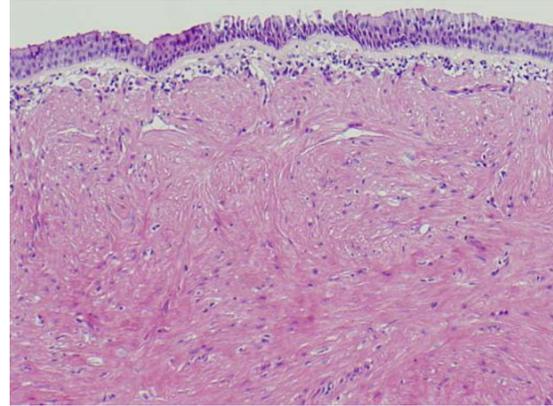
Repeat bronchoscopy 3 months later revealed a 1.5 mm nodule with otherwise normal narrow band imaging. The nodule was resected and revealed benign bronchial tissue. On follow-up her symptoms resolved.



**Figure 1. CT scan showing endobronchial tumor in the bronchus intermedius**



**Figure 2. Bronchoscopic appearance of endobronchial tumor before and immediately following cryotherapy**



**Figure 3. Histological appearance of the tumor showing submucosal spindle cell proliferation with bland cytologic features and no increase in mitotic rate. The spindle cells expressed desmin, smooth muscle and muscle specific actin compatible with leiomyoma (See description in the discussion section)**

### Discussion

Endobronchial leiomyomas are rare benign tumors compromising about 2% of the excised benign endobronchial lesions reported in the literature with only 108 cases from 1950 to 2008.<sup>1,2</sup> These tumors are more commonly diagnosed in the third and fourth decades of life, without sex predilection. However, parenchymal leiomyomas are more common in women, likely due to the presence of benign metastasizing uterine leiomyomas in lung specimens. Such leiomyomas can be confused with primary pulmonary leiomyomas, resulting in over reporting of these cases in women.<sup>2</sup>

Symptom severity correlates with the location and degree of airway compromise. Dyspnea, cough, wheezing and recurrent pneumonias are most frequent presentations. Hemoptysis and rare cases of pneumothorax from extreme air trapping have also been reported in the literature.<sup>3</sup> These lesions are identified as hypovascular and have low attenuation on CT analyses (from 25–46 HU on unenhanced CT and 46–85 HU on enhanced CT).<sup>4</sup>

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Treatment has traditionally been surgical; including tracheobronchial resection, lobectomy and even pneumonectomy depending of the location, size, width of the base of the tumor, and the extent of the pulmonary change distal to the lesion.<sup>5</sup>

More recent reports suggest that bronchoscopic interventions can provide successful therapy of tumors that are exclusively endoluminal. An “iceberg” tumor growth pattern (a small intraluminal component and a large extraluminal component) more typical of carcinoid and mucoepidermoid carcinomas, is not common in leiomyomas, thus favoring interventional bronchoscopic approaches.<sup>6</sup>

In one review of 10 patients with tracheobronchial leiomyoma, bronchoscopic intervention with rigid bronchoscopy and Nd-YAG laser resection was successful in all but one case, which had a broad base tumor and required surgery. Only one case out of 9 had evidence of recurrence in 16 months, thereby requiring subsequent surgery. Bleeding was not significant and only one case had a significant complication related to a bronchial lymph node fistula that was managed by a temporary silicon stent.<sup>7</sup> Overall, Argon plasma coagulation, electrocautery and Nd-YAG laser have resulted in complete endoscopic resection with a very low risk of recurrence, leading to complete symptom resolution.

Cryotherapy has been used as a treatment modality for endobronchial tumors for palliation and treatment purposes. It involves repeated cycles of freezing with temperature usually  $-30^{\circ}\text{C}$  to  $-40^{\circ}\text{C}$  and sometimes reaching down to  $-100^{\circ}\text{C}$  at the tip of the flexible probe being introduced through the flexible or rigid bronchoscope. Repeated cycles of freezing cause necrosis of the tissue and thrombosis of the blood vessels, decreasing associated complications, as

compared to other thermal injury approaches (e.g. laser therapy).<sup>8</sup> Benefits of cryotherapy include low cost, a more controlled approach at tissue destruction with decreased risk of airway perforation (related to not affecting the bronchus cartilage or collagen), less bleeding risk and no risk of airway fire. The latter allows use of higher fractions of oxygen supplementation during the intervention. A systemic literature review by Lee et al<sup>9</sup> based on 16 studies from 1981 to 2008 using cryotherapy for treatment of lung and bronchial tumors found that the overall complication rate (including hemorrhage, mediastinal emphysema, atrial fibrillation and dyspnea) occurred in 0-11.1% of the cases, with these being controlled with simple conservative treatments. These cases were, however, from mostly inoperable and advanced lung cancers rather than the benign lesions.

Historically, cryotherapy was not recognized as an appropriate method for rapid re-establishment of airway patency. This related to the need for repeated sessions to allow the bulk of tumor to undergo necrosis and slough off at a later time, therefore having a delayed effect as compared to other interventions such as laser therapy. However, use of “cryo-recanalization techniques” in recent years using newer flexible probes with effective freezing and penetration has allowed for tumor de-bulking through a freeze-pull method, without the need of repeated sessions to re-establish airway patency.<sup>10</sup>

### Conclusion

Endobronchial leiomyoma is one of the rare benign tumors of the airway. Bronchoscopic resection should be considered as the initial preferred approach although surgery may be required for some cases. Given the benign nature of luminal leiomyomas, cryotherapy can be performed

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for endoscopic resection of these tumors along with good control of local bleeding. It will result in immediate relief of obstruction with minimal complications, as compared to interventions utilizing argon plasma coagulation, laser therapy or electrocautery. To our knowledge, there are no reports using cryotherapy for complete resection of benign endobronchial leiomyomas. This modality should be considered as first line therapy , with ongoing bronchoscopy surveillance to re-evaluate for long term recurrence.

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