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Case Report

Successful Resection of an Endobronchial Pleomorphic Adenoma Using Cryotherapy: A Case Report

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Abstract

Background
Pleomorphic adenomas are very rare benign bronchial tumors. They are typically either removed surgically, or if not amenable to surgery, via therapeutic bronchoscopy. We present the first case of an endobronchial pleomorphic adenoma removed by cryotherapy.

Case Report
This is the case of a 54-year-old man with a right middle lobe pleomorphic adenoma that was detected incidentally during a bronchoscopy, which was performed due to acute respiratory failure and presence of ground glass opacities on imaging. The tumor led to a chronic cough and partial right middle lobe atelectasis. It was resected successfully using endobronchial cryotherapy without any noted complications. There was no recurrence of the disease one year after the intervention.

Conclusion
This is the first report in the literature of a pleomorphic adenoma successfully removed via cryotherapy without recurrence. Endobronchial cryotherapy appears to be an effective and safe method of removing endobronchial pleomorphic adenomas.

Keywords: Pleomorphic adenoma; Cryotherapy; Bronchoscopy; Bronchial Neoplasms; Case report

Introduction

Primary pleomorphic adenomas are the most common tumors of the salivary glands. They can rarely present as benign lesions in the proximal airways and lung parenchyma. In limited case reports, pulmonary pleomorphic adenomas have been removed using electrocautery snare, argon plasma coagulation, and surgical resection [1-4]. Endobronchial cryotherapy has shown success in the resection of benign and malignant airway tumors, and in the treatment of tracheal and bronchial stenosis [5-7]. We hereby present the first case of successful resection of a pulmonary pleomorphic adenoma using endobronchial cryotherapy.

Case Presentation

A 54-year-old man presented with progressively increasing shortness of breath over several weeks' duration. This was accompanied by worsening of a chronic cough of 6 weeks duration. No fever was reported. The patient underwent a renal transplant more than 10 years ago, and had stage II chronic kidney disease upon presentation. He had a history of gastric lymphoma for which he underwent eradication therapy for *H. pylori*. He also had a history of papillary thyroid cancer for which he underwent total thyroidectomy. Both his gastric lymphoma and thyroid cancer were in remission for the last five years. His medications consisted of a proton pump inhibitor, mycophenolate mofetil and prednisone. He was a non-smoker.

In the emergency department, the computed tomography scan of the chest showed bilateral multilobar ground glass infiltrates. Due to worsening of his respiratory status, the patient was admitted to the intensive care unit where he was intubated and remained so for seven days. A diagnostic bronchoscopy was then performed and a shiny, well demarcated mass was noted. The mass was partially occluding the lateral subsegment of the right middle lobe bronchus. Bronchoalveolar lavage of the right middle lobe showed lymphocyte-predominant lavage fluid. Endobronchial biopsies of the mass were consistent with pleomorphic adenoma. He was treated with prednisone 1 mg/kg oral daily and antibiotics. The patient did improve and was later discharged stable from the hospital. After being lost to follow up for one year, a repeat CT scan of the chest was done and showed that the mass in the right middle lobe had enlarged significantly (Figure 1a). A repeat bronchoscopy showed the tumor occluding the lateral subsegment of the right middle lobe bronchus (Figure 1b).

Due to the patient's comorbidities and high-risk status, a decision was made for endobronchial debulking of the tumor. The tumor was debulked using cryoadherence, whereby the 2.4 mm cryoprobe adhered to the tumor and freezing was done for 4 seconds without thawing. This was done multiple times in order to remove most of the tumor (Figures 2a,2b,2c).

Finally, freezing of the residual tumor was done for 1 minute without thawing to hasten cell death. Pathology of the debulked tumor confirmed bronchial pleomorphic adenoma (Figure 3). The patient's cough resolved completely within one week of the procedure. Repeat bronchoscopy done 6 weeks afterwards revealed complete macroscopic resolution of the tumor (Figure 4). The tumor did not recur after more than one year of follow-up.

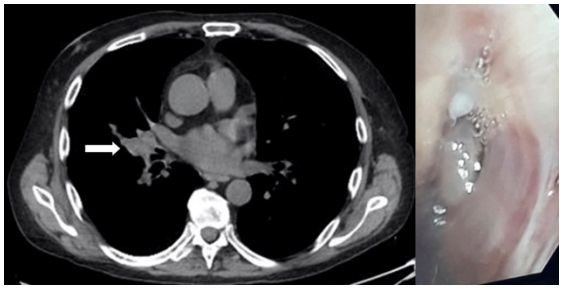

Figure 1: (a) Computed tomography scan of the chest showing the pleomorphic adenoma occluding the lateral subsegment of the right middle lobe bronchus with post-obstructive atelectasis. (b) Bronchoscopic image of the pleomorphic adenoma at the bifurcation of the medial and lateral subsegments of the right middle lobe.

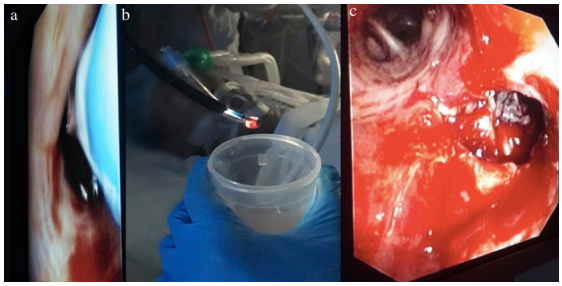

Figure 2: (a) The 2.4 mm cryoprobe can be seen freezing the right middle lobe mass (b) The cryoprobe removed from the airway after freezing without thawing. A piece of the adenoma is seen attached to it (c) The lateral subsegment of the right middle lobe at the end of the procedure. A residual pleomorphic adenoma is seen medially

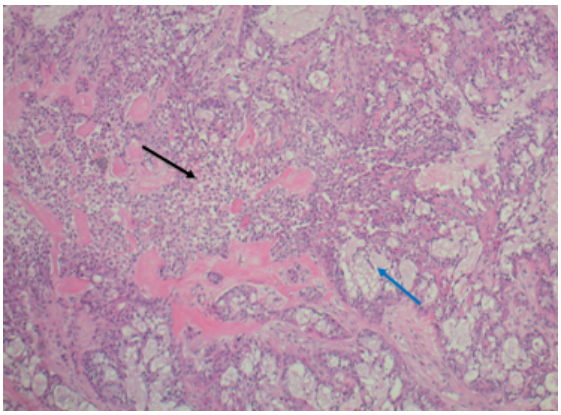

Figure 3: Pathology of the lung mass confirming the diagnosis of pleomorphic adenoma. The blue arrow points to parenchymatous glandular cells of pleomorphic adenoma. The black arrow points to the myoepithelial component of the adenoma.

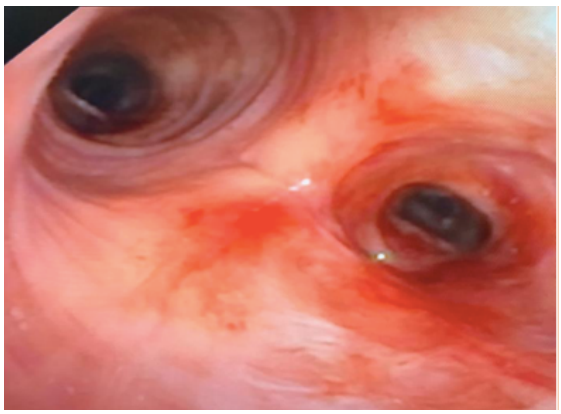

Figure 4: Bronchoscopy six weeks afterwards showing complete macroscopic resolution of the pleomorphic adenoma.

Discussion

Pure tracheobronchial tumors are rare tumors and represent only 0.6% of all pulmonary tumors. Most of them are malignant, while benign tracheobronchial tumors are very rare and include hamartomas, lipomas, papillomas, among others. Pleomorphic adenoma is one of the rarest types of benign tracheobronchial tumors [8]. They are usually asymptomatic, but can grow in size large enough to occlude the airway leading to symptoms. These tumors can rarely metastasize after many years, and therefore when discovered, should be removed either surgically or by therapeutic bronchoscopy if surgery is high risk [9]. Other salivary gland tumors found in the lungs include adenoid cystic carcinomas, which are most centrally located, as well as mucoepidermoid carcinomas, which are more distal. These tumors tend to present with an obstructive pattern (pneumonia, atelectasis). They occur more frequently in younger patients and their prevalence is 6% of all head and neck tumors and less than 2% of lung cancers [8]. It is almost impossible to differentiate between them clinically. They have better prognosis than more common lung cancer types and a 5- to 10-year survival rate of 65 and 53%, respectively [8].

Usually, we opt for surgical resection with possible radiotherapy as the rate of recurrence of pleomorphic adenoma is high. If surgery is contraindicated, then endoscopic resection is performed. As in this case, immunosuppressed status is a risk factor for the development of this type of tumor with an aggressive behavior. This could possibly have been part of an underlying multiglandular autoimmune diseases but the patient was never worked up for this.

Therapeutic bronchoscopic methods that have been used in the limited case reports published in the literature include argon plasma coagulation and electrocautery snare [1-4]. Endobronchial cryotherapy has shown success in the treatment of malignant and benign airway tumors with improved symptoms and removal of these tumors. It has also shown success in the management of benign airway stenosis with removal of granulation tissue and delay of recurrent airway stenosis. It is

considered to be a safe method as it does not affect cartilage, and therefore the risk of airway perforation is extremely low. The cryoprobe utilizes nitrous oxide gas which freezes to below -40° Celsius. Once the probe adheres to the tumor, it leads to the formation of extracellular and intracellular crystals, which results in cellular dehydration, membrane disruption, and death of intracellular organelles such as the mitochondria and endoplasmic reticulum. Endobronchial cryotherapy works by means of cryoadherence, in which the probe adheres to the tumor. Rapid freezing (4 seconds) without thawing can lead to tumor attachment to the probe and immediate removal of the tumor pieces, thus preserving architecture and diagnostic accuracy, as well as allowing for endobronchial debulking. Prolonged freezing (30 seconds to 1 minute) leads to cellular dehydration and cell death as described above and delayed removal of the tumor (1-2 weeks) [5-7].

The differential diagnosis of benign endobronchial tumors includes endobronchial pleomorphic adenoma, endobronchial neurogenic tumor/schwannoma, squamous papilloma, endobronchial mucous gland adenoma, and glandular papilloma among others.

Conclusion

To our knowledge, we here describe the first case report of removal of a bronchial pleomorphic adenoma using endobronchial cryotherapy. This removal utilized the two techniques of immediate tumor removal via rapid freezing without thawing, and delayed removal of the residual tumor via prolonged freezing with slow thawing or warming. This case report shows that this technique may be an effective and safe technique for the treatment of airway pleomorphic adenomas not amenable to surgery.

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