

Endobronchial Argon Plasma Coagulation for Treatment of Dieulafoy Disease

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Dieulafoy disease of the bronchus is a rare condition. A 28-y-old male patient with recurrent exacerbations of hemoptysis (producing up to 300 mL) over the previous 4 y was admitted to our hospital following an acute episode. A 3-mm sessile lesion covered with somewhat ulcerated smooth mucosa was detected at the distal end of the left main bronchus, at the entrance to the upper lobe bronchus. Bleeding was controlled by argon plasma coagulation, and total coagulation of the lesion was also therapeutic. The patient has been followed for 2 y with no further bleeding. In the very few reported cases of Dieulafoy disease, surgical resection and selective embolization were the treatments used. Endobronchial treatment has not been described before as a therapeutic approach. We emphasize that argon plasma coagulation can be a treatment option in such cases. Key words: Dieulafoy disease; bronchoscopy; coagulation; hemoptysis; vascular malformation. [Respir Care 2015;60(1):1–•. © 2015 Daedalus Enterprises]

Introduction

Dieulafoy disease was first described by the French physician Dieulafoy as a vascular malformation of the stomach.¹ Later, it was also defined as a rare disease of the bronchial system. The disease appears with massive and submassive hemoptysis episodes.² On bronchoscopy, a millimeter-sized lesion covered by smooth mucosa is observed. If a lesion is detected between bleeding exacerbations, a biopsy can result in catastrophic hemorrhage.³ Experience regarding its treatment is limited to a few case reports. Surgical resection in a few cases and selective embolization in others have been used as treatments.^{4,5} To

the best of our knowledge, endobronchial treatment to stop bleeding episodes has not been reported before. Argon plasma coagulation is a safe method for the treatment of visible hemorrhagic lesions of the tracheobronchial tree. In this case report, a lesion located at the entrance to the left upper lobe was treated by argon plasma coagulation, and no bleeding episode was observed in the following 2 y.

Case Report

A 28-y-old male patient presented with recurrent hemoptysis, sometimes with massive amounts, over the previous 4 y. The first massive hemoptysis episode occurred in 2008 and again in 2009 and 2012, with the last one producing ~300 mL. The patient had a 1 pack-year smoking history and smoked for 5 y. Other systemic examinations and laboratory values were unremarkable. No abnormality was detected in his thoracic computed tomography scan, with no infiltration. Ground-glass areas due to aspiration of the blood were seen (Fig. 1). Although bronchoscopy had been performed several times at different centers, no lesion had been detected. In another bronchoscopy, performed in our unit following a massive hemoptysis episode, a 3-mm sessile lesion was observed at the entrance to the left upper lobe bronchus and was evaluated as a vascular malformation (Fig. 2). As in other Dieulafoy

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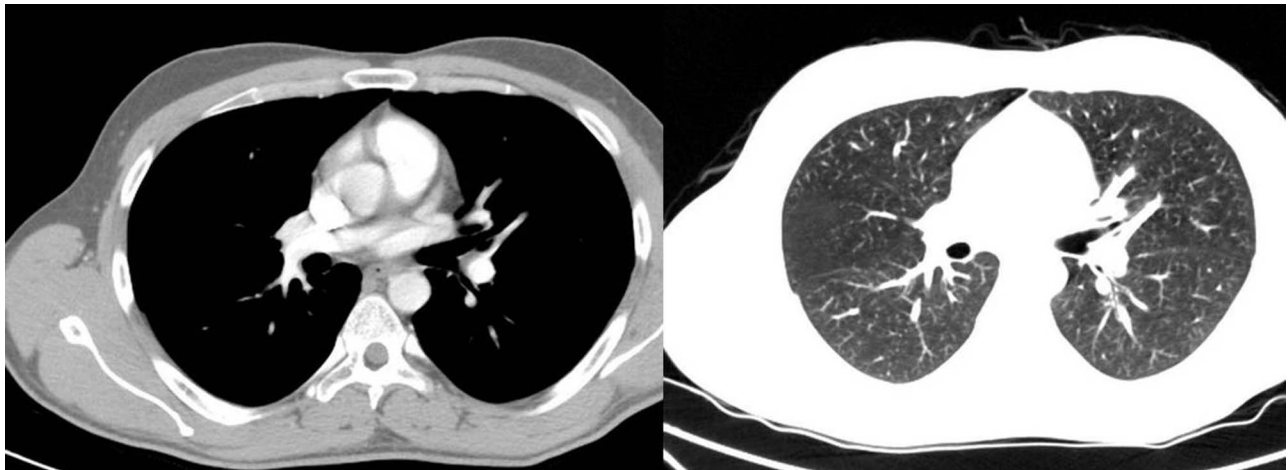


Fig. 1. Thoracic computed tomography sections. There is no remarkable lesion that can explain the hemoptysis.

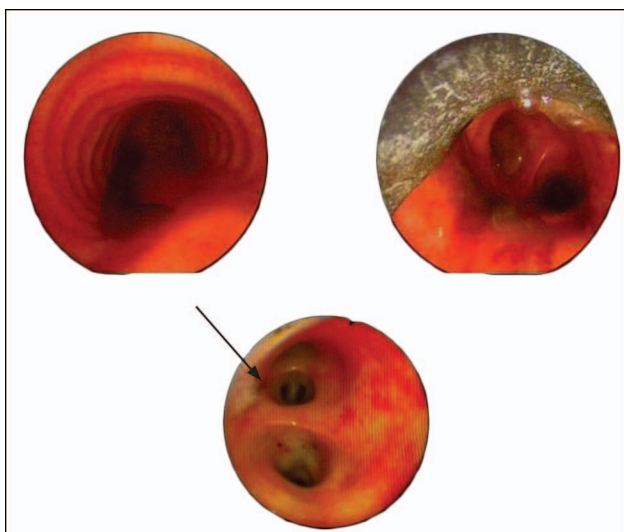


Fig. 2. Rigid bronchoscopy revealed a large coagulum in the trachea. After bronchoscopic cleaning, a small lesion could be seen at the entrance to the left upper lobe (arrow).

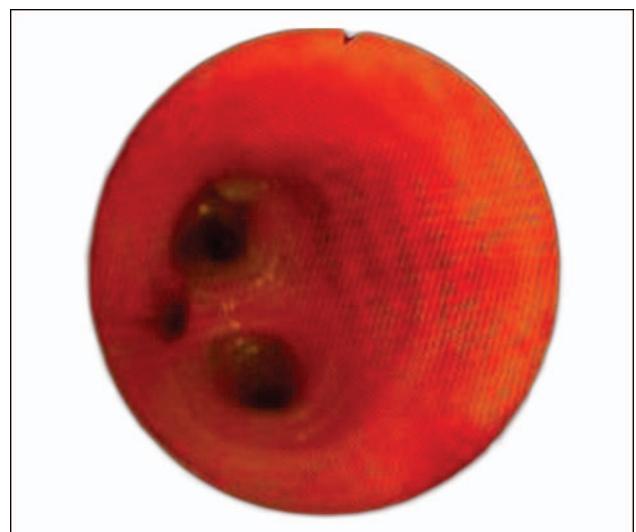


Fig. 3. The lesion was coagulated and carbonized by argon plasma coagulation during the same session in which the lesion was visualized.

disease cases, the lesion was sessile and covered by mucosa that was partly ulcerated. The previous procedures had been performed using rigid bronchoscopy. However, when a flexible bronchoscope was passed through the rigid bronchoscope, the lesion was reached properly. The power setting during argon plasma coagulation was 40 W, and the continuous mode was used. Using a flexible argon plasma coagulation probe, the base and then the surface of the lesion were coagulated (Fig. 3). The total duration of the procedure was 20 min. In a follow-up bronchoscopy performed 10 d later, the lesion had resolved, and no other lesion was observed. The patient did not agree to any further investigation, such as selective bronchial angiography (which can show abnormal bronchial artery forma-

tion) or gastrointestinal endoscopic imaging (to assess any other vascular malformation in the gastrointestinal tract).

There has been controversy regarding the diagnosis of Dieulafoy disease of the bronchus. However, there are no well-defined criteria to establish a diagnosis of bronchial Dieulafoy disease because of its rarity. Nonetheless, the lack of selective bronchial angiography is accepted as a limitation for a definitive diagnosis. In the present case, the lesion at the entrance to the left upper lobe was clearly vascular in nature, and the result of the therapy confirmed the diagnosis. The patient's medical history did not include any gastrointestinal bleeding. No acute hemoptysis was observed in the 2 y after argon plasma coagulation, and the patient is still being followed.

Discussion

Dieulafoy disease was first defined in the gastrointestinal system. Any part of the gastrointestinal system can be affected.⁶ Dieulafoy disease of the bronchus is a rare disease. Very few cases have been reported in English medical literature.^{3,4} The small number of cases may be partly due to the fact that it is difficult to detect the lesion bronchoscopically, and the lesion may be missed even when several bronchoscopies are done, as in this case.

Surgical treatments have been reported in some cases, and in others, good results have been obtained with embolization. To the best of our knowledge, no case treated by an endobronchial approach, including the gastrointestinal tract, has been reported.⁶ Argon plasma coagulation has a lower penetration depth and is selectively oriented to the bleeding area; it is a frequently used method for visible hemorrhagic lesions of the tracheobronchial tree. Although the use of argon plasma coagulation is generally safe, a major complication can be air emboli.⁷ Unlike laser use, argon plasma coagulation does not cause serious complications, such as perforations or major secondary bleeding. For this reason, it is frequently used in connection with mechanical resections of malignant obstructive lesions and benign tumors of the airways. It can also be used safely for eradication of basal areas of resected tumors.

The efficacy of bronchoscopic laser treatments of vascular malformations has long been known, since the first endobronchial laser applications.⁸ However, argon plasma coagulation was not considered a treatment option for these patients. Coagulation of the whole surface, starting from the basal area of the lesion, causes shrinkage of the lesion. In this way, the bleeding point is covered. In the healing phase, significant scars in the submucosal layer provide long-term bleeding control. In this case, one application of argon plasma coagulation was sufficient for long-term bleeding control. The lesion resolved, and no stricture remained in a follow-up bronchoscopic exam in the healing

phase. The extension of the bronchus through the submucosal layer can help to coagulate a widened bronchial artery with this method. In some cases, more than one lesion has been seen at the mucosa. It may be more appropriate to use embolization or surgery as the treatment option rather than argon plasma coagulation in such cases.

Endobronchial Dieulafoy disease is a rarely seen entity. Due to their small sizes and covered with coagulum, associated lesions may be rarely recognized, and this could have fatal results. In such cases with no apparent lesion, a very careful bronchoscopic evaluation is essential. In conclusion, in addition to successful treatment options, such as surgery and selective embolization, argon plasma coagulation should also be kept in mind as a safe and effective method.

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