Case Report

Diffuse Endobronchial Telangiectasia

Abstract

Hemoptysis is one of the most common reasons for seeking emergency care. Infections and malignancy are the leading causes of hemoptysis although caused by various other pulmonary and extrapulmonary conditions. Most causes are self-limiting and do not warrant any aggressive investigation. Endobronchial telangiectasia can rarely cause hemoptysis and is seen in patients with hemorrhagic hereditary telangiectasia or scleroderma. Isolated diffuse endobronchial telangiectasia is rare and is only reported in one case in literature. We present another case of diffuse endobronchial telangiectasia in a young adult who presented with recurrent hemoptysis. Computer tomography scan was normal, but bronchoscopy showed multiple endobronchial arteriovenous malformations in the entire tracheobronchial tree.

Keywords: *Arteriovenous malformations, endobronchial telangiectasia, hemoptysis*

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Introduction

Hemoptysis is defined as the coughing of blood arising from the lower respiratory tract. It is one of the common reasons for seeking emergency care in adults. As blood in the airway can clot and can compromise of airway patency, hemoptysis is the most feared of all the pulmonary symptoms. Bronchial infections and malignancies are the two most common causes of hemoptysis throughout the world and can be seen in a variety of pulmonary and systemic disorders. Most cases are self-limiting and do not need further investigation. Computer tomography (CT) scan of the chest can detect most causes of hemoptysis and is most commonly used for patients presenting with significant hemoptysis. The routine use of bronchoscopy in patients with hemoptysis is still being debated. We present a case of recurrent hemoptysis in a young patient who had normal CT scan of the chest on multiple occasions and was easily diagnosed with the help of bronchoscopy.

Case Report

A 47-year-old male was referred to the pulmonary clinic for the further evaluation of hemoptysis. It started 2 years back when suddenly 1 day he started coughing up blood. It was bright red associated with fresh clots and warranted visit to the emergency room. He denied any shortness of breath, fever, chest trauma, and chest pain before or during the hemoptysis. He also denied a preceding cough before the onset of hemoptysis. CT scan of the chest and routine blood work done in the emergency room were reported to be normal, and he was discharged home with course of antibiotics and inhalers. The hemoptysis eased during the following day, and he only coughed up minimal dark colored sputum for the next few days. It completely subsided in 3–4 days. Since then, the patient had almost 12 similar episodes of hemoptysis, once every 3–4 months and lasting for less than a week. He denied any predisposing factors for his symptom and has not noticed any postural or diurnal variation. The patient also categorically denied any respiratory problems between these episodes. He does not take any prescription or over-the-counter medicines. He smoked for a few years during his college days and quits smoking almost 15 years back. The patient had four emergency room visits so far, and multiple CT scans of his chest and extensive blood investigations were unrevealing. During the last visit, he also underwent upper airway evaluation by laryngoscope and was reported to be normal. Although he was referred to see pulmonologist before, he could not keep the appointment due to lack of health insurance.

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To evaluate further the cause of recurrent hemoptysis, diagnostic bronchoscopy under moderate sedation was done through the right nostril. The upper trachea was normal. However, beginning in the lower trachea and extending beyond the visualized subsegmental bronchi, there were diffuse 1–2 mm multiple telangiectasias noted in both side airways [Figure 1]. None of them were obviously bleeding. His last hemoptysis was 3 weeks back. On further questioning, he denied any epistaxis, gastrointestinal bleeding, or cutaneous telangiectasia. Upper and lower gastrointestinal endoscopy did not reveal any arteriovenous (AV) malformation. Transthoracic echocardiogram with contrast did not reveal any right-to-left shunt. Workup for collagen vascular disease including scleroderma was negative. Based on the available information, the patient was diagnosed with diffuse endobronchial telangiectasia. The treatment options for endobronchial telangiectasia were discussed with the patient. The limitation of endobronchial treatment in his case due to the diffuse nature of the disease were also explained in detail. The patient missed his follow-up in the pulmonary clinic. He cited lack of insurance and financial reasons for missing appointments. He did one more episode of hemoptysis which was once again self-subsiding.

Discussion

Sudden-onset hemoptysis can be frightening and also life-threatening. Patients presenting with massive hemoptysis need stabilization of airway. The management strategies for patients presenting with nonmassive hemoptysis can be challenging. Both CT scan of the chest and bronchoscopy are used for identifying the cause of hemoptysis, and each one has its own merits. However, the optimal use of both these investigations is still being debated. CT scan of the chest is less invasive and can be done timely. CT scan of the chest done during active hemoptysis is known to detect most causes of hemoptysis. Bronchoscopy can be complementary to CT scan in detecting endobronchial lesions and also site of lesions.[1,2] With the growing arsenal of endobronchial interventional technology, flexible bronchoscopy is also therapeutic in most cases.

Isolated telangiectasia seen in the endobronchial tree is rare and only reported a handful of cases.[3-7] Hereditary hemorrhagic telangiectasia (HHT) is an autosomal dominant vascular disorder developmental of small AV malformations in multiple organs including lungs. Telangiectasia can develop in the endobronchial tree and can cause hemoptysis.[4] Endobronchial telangiectasia can also be seen in patients with scleroderma.[5,6] However, isolated endobronchial telangiectasia is only described in one case report in the literature.[7] In 1974, Masson *et al*. described a patient with multiple flat, red, nonpulsatile areas of tiny superficial vessels in a patient with long-standing hemoptysis. As endobronchial telangiectasia is not seen without the stigmata of HHT, the author concluded this to be congenital aberration of the bronchial vasculature.

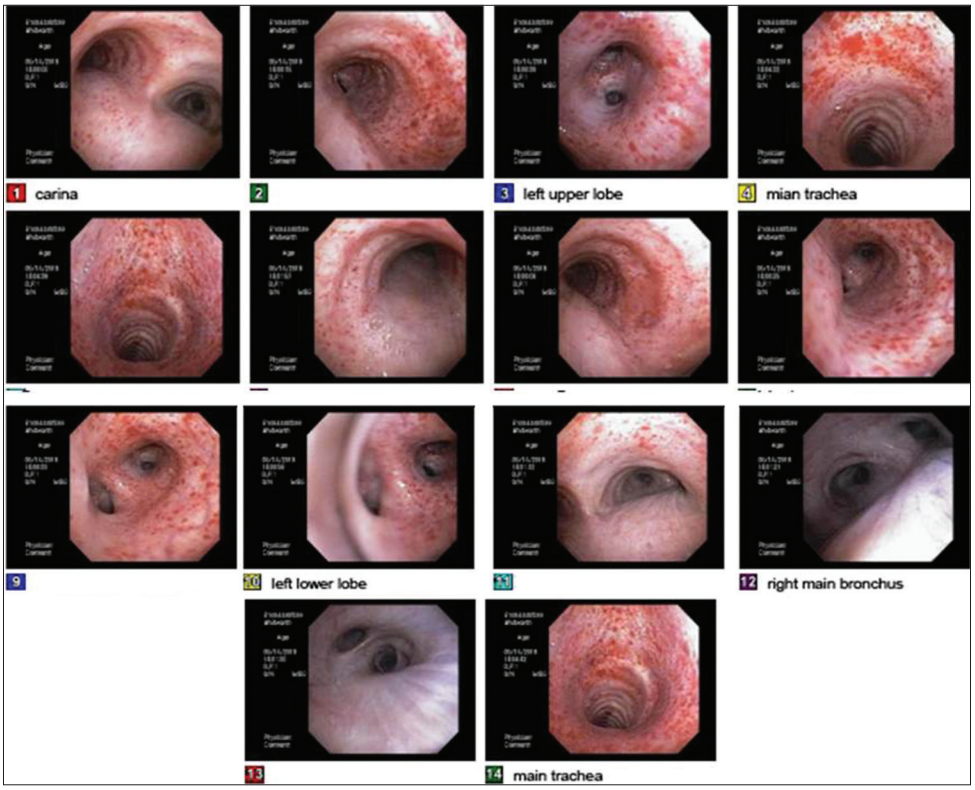

Figure 1: Bronchoscopic images showing diffuse endobronchial telangiectasia

Our patient did not show any features consistent with HHT such as epistasis, skin lesions, or family history. Due to financial constraints, he declined to follow-up with HHT center nor got the genetic workup for that. There was no clinical or serological evidence of scleroderma. We also believe that multiple AV malfunctions seen in all patients are likely a developmental disorder. Endobronchial laser therapy and surgical resections have been successfully used to treat endobronchial telangiectasia. However, diffuse and bilateral involvement in our patients precluded these.

Conclusion

Diffuse endobronchial telangiectasia is rare. It can cause nonmassive recurrent hemoptysis in adults. CT of the chest is normal in this case and is diagnosed by diagnostic bronchoscopy. Once again, our case illuminates the utility of bronchoscopy in patients with nonmassive recurrent hemoptysis and has normal CT scan of the chest.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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