An Unexpected Cause of Hemoptysis: Endobronchial Lipomatous Hamartoma

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ABSTRACT
Hamartomas are the most common benign tumors of the lung. Endobronchial hamartomas are even rarer and infrequently cause hemoptysis. We report a case of endobronchial hamartoma that was originating from a segment bronchus and invisible in chest X-ray. A 63-year-old man was admitted to hospital with hemoptysis. A CT scan revealed endobronchial mass obstructing anterior bronchus of the right lower lob of the right lung. It wasn’t radiographically presented. Flexible bronchoscopy detected a polypoid mass (1.5x1.0 cm) that arising from the posterior wall of the anterior segment of right lower lob. Histopathologic examination revealed lipomatous hamartoma. It was resected with an electro-surgical snare. Cryotherapy was applied to residual lesion on surface of the bronchus. The patient was successfully recovered. In conclusion, lipomatous hamartoma may presented as rare cause of hemoptysis. Endoscopic treatment is safe and currently modality used for select cases.

Key words: Endobronchial hamartoma, hemoptysis, cryotherapy.

1. INTRODUCTION
Pulmonary hamartoma described common benign lung tumor with an incidence of 0.025-0.32% (1). There are two clinical type as location: intraparenchymal or endobronchial. Parenchymal hamartomas are usually asymptomatic and presenting as solitary pulmonary nodule. Endobronchial hamartomas originate from large bronchus and grow into the lumen. It may result atelectasis or pneumonia. Common symptoms include such as cough, dyspnea, wheezing (1, 2, 3). It may cause misdiagnoses as asthma (4). Hemoptysis can occur, but it is unusual. Endobronchial lipomatous hamartomas are not vascularized, therefore they are not typically associated with hemoptysis (5).

CASE REPORT
63-year-old man was admitted to the hospital with the complaints of hemoptysis. He reported that there was a little bloody sputum intermittently for 10 days. There was no an additional symptoms. He had never smoked. The patient had been admitted to nearly hospital 10 days ago and had been told that his chest radiography and CT scan had revealed no abnormal findings. Physical examination revealed a healthy looking man. Chest auscultation was normal and vital findings were stable. Laboratory evaluation included hemoglobin of 14.4 g/dl, white blood cell count of 5.400 with normal platelet count and coagulation profile. Erythrocyte sedimentation rate was 14 mm/h. Renal and liver function tests and chest radiography (Figure 1) were normal. A CT scan revealed a low attenuation endobronchial mass obstructing the anterior bronchus of the right lower lob (Figure 2). The bronchoscopy was performed under local anesthesia. Flexible bronchoscopy detected a smooth surface with a wide sessile base polypoid lesion partially occluding the right lower lob anterior segment bronchus (Figure 3). Endobronchial biopsy was obtained from the lesion. It was diagnosed as a lipomatous hamartoma based on the bronchoscopic biopsy. On second bronchoscopy, the hamartoma was resected with an electro-surgical snare. Cryotherapy was applied to residual lesion on surface of the bronchus. The excised material consisted of gray, shine mucosal tissue, 1.5 x 1.0 cm in diameter. The histologic examination of the tumor was confirmed as lipomatous hamartoma (Figure 4). No complications occurred. One month later, on following fiberoptic bronchoscopy the lower lob anterior bronchus was completely open and no residual tissue. Hemoptysis did not recur.

2. DISCUSSION
Hamartomas originate from bronchial wall mesenchymal tissue and involve cartilage, adipose, fibrous and epithelial elements. The peak incidence is in the seventh decades of life, they
Figure 2. Chest CT shows a low attenuation endobronchial mass obstructing the anterior bronchus of the right lower lob (coronal view) are more common in males (6). They are usually small and well demarcated. Average size is 1.5 cm, although some are as big as 6 cm (6). In our case, it was 1.5 cm and well circumscribed.

They are generally asymptomatic because of peripheral location and accidentally found in chest radiography routine evaluation. Symptoms of endobronchial hamartoma develop due to obstruction of the tracheobronchial lumen, as fever, cough, expectoration, wheezing, and dyspnea (2, 4). Endobronchial tumor may be discovered by accidentally during investigating other respiratory or cardiac diseases (3, 5, 6, 7). Lipomatous form are not vascularized and thus don’t usually cause hemoptysis (5). Our case was diagnosed lipomatous hamartoma and single symptom was hemoptysis. It may represent impingement of the hamartoma on adjacent vascular structures. Massive hemoptysis reported only tree cases in literature (8, 9, 10). Conventional radiography is often nonspecific and related to post-obstructive changes, such as atelectasia, pneumonia, bronchiectasis (11). In parenchymal hamartoma, the typical radiographic appearance is in the form of solitary pulmonary nodule that it may contain popcorn calcification. However, endobronchial mass lesion may be poorly demonstrated or not shown at all on chest radiography (12). Endobronchial hamartoma usually appears at CT collections of fat alternating with foci of calcification. It typically contain more fat tissue than parenchymal ones (11). In our case, chest radiography was normal. A low attenuation endobronchial mass in peripheral bronchus was detected on CT but the lesion wasn't seen on axial CT image. Therefore, this lesion couldn’t be realized when the patient went first. We only saw it on coronal CT image and it contained fat density. The tumor in this case was visible on bronchoscopy and histopathologically verified lipomatous hamartoma.

Pulmonary hamartomas have little malignant potential but they may grow. Thus, they should be treated. Surgical resection is a traditional treatment for these tumors. Bronchoscopy is a modality currently used for the diagnosis and treatment. Various bronchoscopic techniques are used for benign endobronchial tumors such as YAG laser, electoacuter, cryotherapy, argon plasma coagulation (13). Endobronchial electrosurgery is inexpensive and easy to perform for the complete resection. Complications of this procedure include hemoptysis, perforation and burning on the tracheal wall. In our case, we excised the lesion by electrosurgery and cryotherapy. The patient did not develop any complications and has remained stable through follow up. Endobronchial hamartomas are usually originating from large bronchus but in this case it was located in segment bronchus. Early diagnosis and treatment received thanks to hemoptysis.

In conclusion, endobronchial hamartoma is uncommon clinic entity and infrequently presents as hemoptysis. It may not seen on chest radiography. If there are findings of bronchi obstruction, such as atelectazia, pneumonia, it can be detected earlier. A CT scan is usually detected as low attenuation endobronchial mass. Endobronchial obstruction by a tumor may cause leading to irreversible pulmonary damage. Thus, early diagnosis and treatment is important. Endoscopic treatment is safe and effective therapeutic option for select cases.

CONFLICT OF INTEREST: NONE DECLARED

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