CASE REPORT

Endobronchial hamartoma: a rare disease with more common presentation

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SUMMARY

Hamartomas are the most common benign tumours of the lung with an overall incidence of 0.025–0.32%. They are known to be amalgamation of various tissue types, originating from the embryonic mesoderm. Lung hamartomas typically involve lung parenchyma and only infrequently grows as endobronchial tumours. We present a case of an 80-year-old man who presented to the pulmonary clinic for consultation for breathlessness, recurrent pneumonias and an abnormal radiograph finding. CT scan of the chest showed scattered infiltrates and atelectasis in the left upper lobe. He underwent a diagnostic bronchoscopy that showed a lobulated endobronchial lesion obstructing the left upper lobe bronchus. Brush and forceps biopsies were obtained that were consistent with an endobronchial hamartoma. He was referred to the pulmonology department for endobronchial debulking of the lesion following which he improved clinically during postoperative follow-up.

BACKGROUND

Malignant tumours are by far the most common lung tumours, accounting for 98% of all lung masses.¹ Among benign tumours, hamartomas are the most common, with an overall incidence of 0.025–0.32%.^{2–3} The majority of lung hamartomas involve the lung parenchyma and only infrequently grow as endobronchial tumours.^{3–5} These tumours occur more frequently in men^{3 6–11} and usually in the elderly.^{4 6 12} Even though the majority of patients will have an abnormality on a chest radiograph, lung hamartomas are usually asymptomatic.^{4 12–15} Obstructive pulmonary symptoms have been reported.^{12 13} CT scans of the chest and bronchoscopy are useful diagnostic tools.^{2 3 7 11 16} Finally, more than one treatment modality has been described in the treatment of pulmonary hamartomas including endobronchial tumours.^{6 7 12 16 17} Among these modalities, rigid bronchoscopy with laser therapy is considered the gold standard, especially for large tumours, and it has replaced the more invasive surgical options.⁷

CASE PRESENTATION

Our patient is an 80-year-old man with medical history significant for coronary artery disease, chronic atrial fibrillation and essential hypertension. He is an ex-smoker who quit 10 years ago with an 80 pack year smoking history. He is an everyday alcohol user of about 12 ounces of regular beer. He worked in refrigeration repair and specifically denied any illicit drug use or exposure history.

The patient was seen initially in the pulmonary clinic for recurrent pneumonias, breathlessness and an abnormal radiograph finding. He reported that he had been diagnosed with pneumonia three times in the last 12 months prior to his visit. His CT scan of the chest ([figure 1](#)) showed persistent scattered infiltrate and atelectasis in the left upper lobe, the latter was more prominent in the lingula with interval increase in diffuse mucous plugging involving the segmental and subsegmental bronchial branches compared to a previous CT from 5 months ago. Given his abnormal chest CT along with his recurrent pneumonias, diagnostic bronchoscopy was performed and showed a lobulated endobronchial lesion obstructing the left upper lobe bronchus ([figure 2A](#)). Brush and forceps biopsies were obtained from the left upper lobe endobronchial mass and were sent for pathology, which showed fragments of benign bronchial mucosa, adipose tissue, submucosal glands, cartilage and myxoid stroma consistent with a pulmonary hamartoma. The overlying bronchial mucosa showed no evidence of dysplasia or malignancy ([figure 3](#)).

TREATMENT

Given the fact that the patient had recurrent post-obstructive pneumonias due to his endobronchial hamartoma, he was referred to interventional pulmonology for endobronchial debulking. In his second flexible bronchoscopy, an exophytic lesion was visualised in the left upper lobe bronchus causing 100% obstruction. The diagnostic scope was unable to be advanced around the lesion. The airways of the left upper lobe and lingula were unable to be seen. The left lower lobe was normal. Electrocautery snare was used to debulk large pieces of the hamartoma. Following removal of the lesion, residual tissue present on the anterior bronchial wall was treated with argon plasma coagulation. Following treatment, there was residual 10% obstruction of the left upper lobe bronchus ([figure 2B](#)). All segments of the left upper lobe and lingula were easily visualised by the end of the procedure.

OUTCOME AND FOLLOW-UP

One week after the procedure, the patient presented in the pulmonary clinic and reported significant improvement breathlessness and no significant residual or new respiratory symptoms.

A follow-up chest CT scan performed 3 months later demonstrated complete resolution of the endobronchial hamartoma and left upper lobe air space disease.

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Figure 1 CT scan of the chest showing an obstructing endobronchial lesion within the main bronchus of the left upper lobe which measures ~17 mm and contains a focal central calcification and fat (A), in addition to persistent diffuse scattered infiltrates and atelectasis throughout the left upper lobe, in addition to extensive progression of mucus plugging (B).

Figure 2 (A) Diagnostic bronchoscopy revealing a lobulated endobronchial lesion obstructing the left upper lobe bronchus. (B) Following treatment, there was residual 10% obstruction of the left upper lobe bronchus. All segments of the left upper lobe and lingula were easily visualised by the end of the procedure.

Figure 3 Pathological evaluation of the brush and forceps biopsies from the left upper lobe endobronchial mass showing fragments of benign bronchial mucosa, adipose tissue, submucosal glands, cartilage and myxoid stroma, consistent with a pulmonary hamartoma. The overlying bronchial mucosa showed no evidence of dysplasia or malignancy.

DISCUSSION

The majority of lung tumours are malignant.¹ Of the small proportion of benign tumours, hamartomas are the most common, with an overall incidence of 0.025–0.32%.^{2,3} Even fewer still are endobronchial hamartomas, with a prevalence ranging from 3% to 12% of all lung hamartomas.^{3–5} Hamartomas are known to be amalgamation of various tissue types, originating from the embryonic mesoderm: predominantly adipose and cartilage, but also bony tissue and smooth muscle.^{6–8} The mass is typically lined by respiratory epithelium.⁷ Unlike malignant tumours, these tissue masses lack cellular atypia⁸ and are not rapidly dividing.⁹ Overall, men are 2–4 times more likely to be affected than women.^{3,6,10,11}

These tumours are most commonly identified between the ages of 50 and 70 years, which is a consequence of their slow growth and generally asymptomatic nature.^{4,6,12} Some studies have demonstrated an increased incidence in smokers.^{1,13} There has also been a suggested association with other tumours as

well, with one study showing 16% of the patients having a concomitant lung cancer,² and another showing 29% of the patients with a secondary malignancy of any type¹¹, though this has not been consistent throughout studies.^{3,4,7}

Since lung hamartomas typically involve the parenchyma, patients are often asymptomatic. However, patients with endobronchial lesions often report symptoms related to airway obstruction, including cough, with or without haemoptysis, fever and dyspnoea. They may have recurrent pneumonias or atelectasis detected on imaging.^{2,12,14} The majority of patients will have an abnormality on chest radiograph,^{4,13,15} though CT may be a better study for endobronchial lesions, whose composition is largely adipose.³ On bronchoscopic examination, these tumours are generally described as smooth, yellow, spongy tissue with an average size of about 1.5 cm.^{2,7,11,15} They do not invade the surrounding tissue but can cause distal destruction of the lung secondary to chronic obstruction.^{2,7,18}

Treatment of these lesions has historically been centred on surgical resection either through wedge resection, lobectomy or pneumonectomy.^{6 12 16} With advances in bronchoscopy, the gold standard for treatment later became rigid bronchoscopy with laser therapy for patients with large lesions.⁷ Unfortunately, many of these patients are still sent for thoracotomy out of concern for malignancy, which clearly carries high operative risk, considering the benign nature of these tumours.¹⁴ There has been a considerable increase in the use of fiberoptic bronchoscopy in recent years for the definitive diagnosis and resection of endobronchial hamartomas,^{1 2 7 17} after which recurrence is considerably rare.^{2 12}

Learning points

- The majority of lung tumours are malignant.
- Endobronchial tumours are not uncommon; however, endobronchial hamartomas are rare.
- Endobronchial hamartomas are the most common benign tumours of the lung. They usually grow inside the lung parenchyma but only infrequently grow as endobronchial tumours.
- With advances in bronchoscopy, the gold standard for treatment has become rigid bronchoscopy with laser therapy especially in patients with large lesions.

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