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Primary pulmonary leiomyoma

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Abstract

Leiomyoma is a smooth muscle neoplasm that commonly occurs in the genitourinary system and the gastrointestinal tract of the body. Primary pulmonary leiomyoma is rarely reported in literature.
We report a rare case of primary pulmonary leiomyoma of a 55-year-old male patient presenting with symptoms of cough for six months.

Introduction

Primary pulmonary leiomyoma is a rare condition that was first reported in 1910. We report a rare case of primary pulmonary leiomyoma of a 55-year-old male patient who presented with symptoms of chronic cough for six months. The lesion was subsequently removed via rigid bronchoscopy.

Case Report

A 55-year-old Chinese gentleman presented to our outpatient clinic with a six-month history of cough, sputum production, and loss of appetite. He also reported a weight loss of 15 kg over one month. Besides hypertension, he had no other past medical history. He was a non-smoker and had not traveled in the last six months; he also did not have any previous tuberculosis or tuberculosis contact. On physical examination, he was comfortable at rest and had no significant physical signs apart from reduced air entry on auscultation at the left lower chest. Blood investigations were unremarkable. His chest radiograph showed a new left lower lobe collapse as compared with previous images. Computed tomography thorax revealed complete collapse of the left lower lobe with subcentimeter mediastinal lymph nodes. An oval endoluminal lesion of size 0.8×1.1 cm was noted in the origin of the left lower lobe bronchus (Fig. 1). Endobronchial ultrasound bronchoscopy was performed. A ball-like endobronchial lesion was seen, causing complete occlusion of the left lower lobe bronchus (Fig. 2). Mediastinal lymph nodes were also seen at 4 and 11 L. Biopsies of the mass and endobronchial ultrasound transbronchial needle aspiration of these lymph nodes were performed. Computed tomography of abdomen, pelvis, and brain showed no evidence of metastatic lesion. An esophageal gastroduodenoscopy was performed, and this was normal. A bone scan was also performed, and there was no conclusive evidence of metastatic disease. Pathological examination of the endobronchial lesion showed a leiomyoma, consisting of interlacing fascicles of spindle and oval cells with eosinophilic and vacuolated cytoplasm, occasional cigar-shaped nuclei, and ill-defined borders. It demonstrated low mitotic activity (0–1 high-power field), with no evidence of diffuse cytological atypia or necrosis. Immunohistochemical staining showed that the tumor cells were strongly positive for smooth muscle markers (smooth muscle actin, desmin, caldesmon) but negative for AE1/3, CD117 (except mast cells), CD34, S-100, HMB45, and synaptophysin. Ki-67 expression is low (1–2%) (Fig. 3).

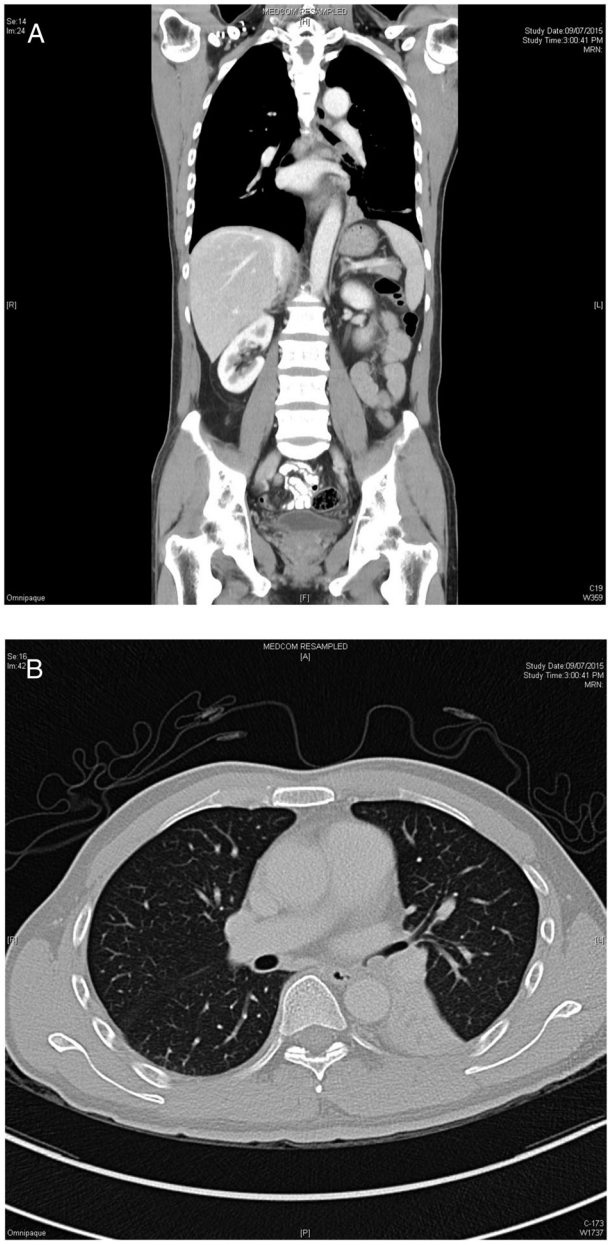

Figure 1. (A and B) Computed tomography (CT) thorax showed oval lesion in left lower lobe bronchus.

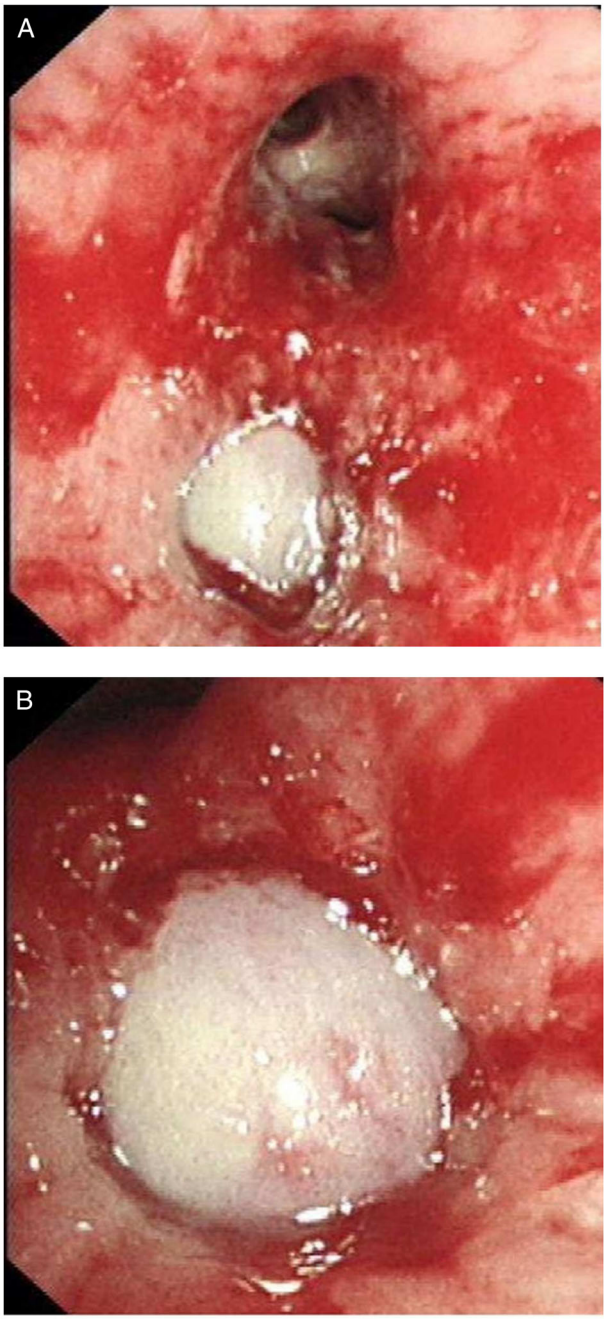

Figure 2. (A and B) Bronchoscopy revealed a ball-like lesion in the opening of left lower lobe.

Pathological result of the two lymph node aspirates showed lymphoid yield without any malignant cells observed. The lesion was located at the left lower lobe bronchus (LB6). The patient subsequently underwent rigid bronchoscopy with complete removal of the endobronchial lesion with cryogenic laser. Post-operative pathological result of the specimen was consistent with leiomyoma. He is on regular respiratory clinic follow-up with an initial follow-up at three months and six-monthly thereafter. He underwent repeat bronchoscopy three months after lesion removal, and there was no evidence of tumor recurrence.

Discussion

Pulmonary leiomyoma is a rare condition with most cases secondary to metastatic pulmonary lesions, from a primary

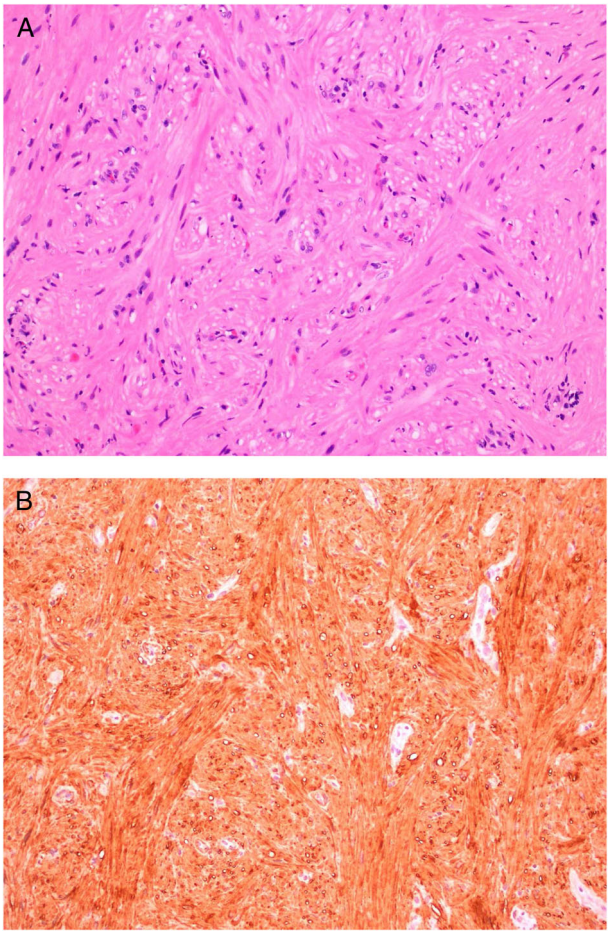

Figure 3. (A) A medium power view demonstrating smooth muscle cells without evidence of atypia or malignancy. There is no cytological atypia, necrosis, increased or atypical mitotic activity (haematoxylin and eosin [H&E] staining, ×200). (B) The spindle cells are strongly and diffusely positive for H-caldesmon, a specific marker for smooth muscle differentiation (H&E staining, ×200).

usually located in the uterus in female patients. Primary pulmonary leiomyoma is scarce, and it usually occurs in parenchymal, endotracheal, or endobronchial sites. Because it was first described by Forkel [1] in 1910, there have been less than 100 reported cases until 2009 [2]. It constitutes less than 2% of all cases of pulmonary benign tumors [3]. Endobronchial leiomyoma accounts for 33–45% of primary pulmonary leiomyoma [3,4].

The nature and etiology of primary pulmonary leiomyoma are still unknown. The diagnosis of primary pulmonary leiomyoma is mainly based on both radiological and pathological investigations. In our case, the normal esophageal gastroduodenoscopy ruled out a gastrointestinal tract origin of leiomyoma. Immunohistochemical stains are useful in ruling out other differential diagnosis; CD117 and CD34 are markers of gastrointestinal stromal tumors; HMB-45 reactivity suggests angiomyolipoma while S-100 usually indicates neural origin. In this case, the immunohistochemical stains of the lesion showed tumor cells that were strongly positive for smooth muscle markers but negative for CD117, CD34, S-100, HMB45, which was consistent with the diagnosis of leiomyoma.

There are currently no guidelines for treating primary pulmonary leiomyoma. The treatment strategy for pulmonary leiomyoma depends on airway location and size of the lesion [5]. Both bronchoscopic resection and surgical resection of the lesion have been reported [2,4,6,7]. In a Korean single institute study reporting 16 cases of primary pulmonary leiomyoma, 11 patients underwent bronchoscopic intervention, of which nine of them had a bronchial leiomyoma, and the other five patients underwent surgical resection of which four of them had tracheal or lung parenchymal leiomyoma [7].

In conclusion, we have reported a rare case of primary pulmonary leiomyoma, which was successfully treated with laser therapy and removal via rigid bronchoscopy.

Disclosure Statements

No conflict of interest declared.
Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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