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Case Report

Peripheral pulmonary artery aneurysm presenting with life-threatening massive hemoptysis: a case report of successful treatment with left upper lobe sleeve resection

Liangchao Wang, Dongping Xia, Xiaoqun Guo, Xiaowu Shi, Yi Hu

Respiratory Medicine, The Central Hospital of Wuhan, Tongji Medical College, Huazhong University of Science and Technology, Wuhan 430014, P. R. China

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Abstract: We report a rare case of peripheral pulmonary artery aneurysm (PPCA) with life-threatening massive hemoptysis. A 62-year-old male presented with repeated cough, serious dyspnea, and massive hemoptysis. He was diagnosed to have chronic obstructive pulmonary disease (COPD) with PPCA on the basis of computed tomography (CT) and high resolution computed tomographical angiography of pulmonary artery. After combined therapy with anti-inflammation, anti-asthmatic, and anti-tussive, pulmonary artery embolization and left upper lobe sleeve resection, the patient recovered smoothly and was followed for more than a year without further recurrence of hemoptysis.

Keywords: PPCA, COPD, dyspnoea, hemoptysis, CT

Introduction

Peripheral pulmonary artery aneurysm (PPCA) is a rare disease [1, 2]. PPCA is associated with vasculitis (Behcet's disease and Hughes-Stovin syndrome) and infectious diseases (tuberculosis, aspergillosis, necrosis, and pneumonia), which may also occur after vessel injury or vessel erosion by a necrotic tumor [3]. Massive hemoptysis is defined as the expectoration of more than 300 ml blood from bronchial tree within 24 hours, which is associated with a mortality rate of over 50% [4, 5]. The majority of massive hemoptysis originates from bleeding hypertrophied bronchial arteries, while less than 10% of massive hemoptysis results from PPCA [6, 7]. In this study, we describe a case report of a 62-year-old man with PPCA as a cause of life-threatening massive hemoptysis.

Case presentation

Informed consent was obtained from all individual participants included in the study.

A 62-year-old man was admitted to our hospital because of repeated cough, serious dyspnea, and massive hemoptysis. The patient complained of progressively increasing dyspnoea of 2-year duration and was diagnosed with chronic obstructive pulmonary disease (COPD) 2-years earlier. The patient gave a history of pulmonary tuberculosis and smoking at the rate of 2 packs per day for 50 years and denied history of disease in the family. General examination showed a pulse rate of 78 beats per minute, a respiratory rate of 22 per minute, and a blood pressure of 125/76 mmHg. Clubbing was present while pallor, icterus, and lymphadenopathy were absent. Respiratory examination revealed bilateral fine end inspiratory rales heard in left upper lobe. Other systems examined were within normal limits. Routine laboratory investigations, including complete blood count, erythrocyte sedimentation rate, blood sugar, hepatic and renal functions were within normal limits. Serum immunological profile revealed antibody to HIV (human immunodeficiency virus) and hepatitis B were negative.

Spirometry could not be performed because of serious dyspnoea and massive hemoptysis. Computed tomography (CT) revealed obsolete pulmonary tuberculosis of bilateral upper lobe

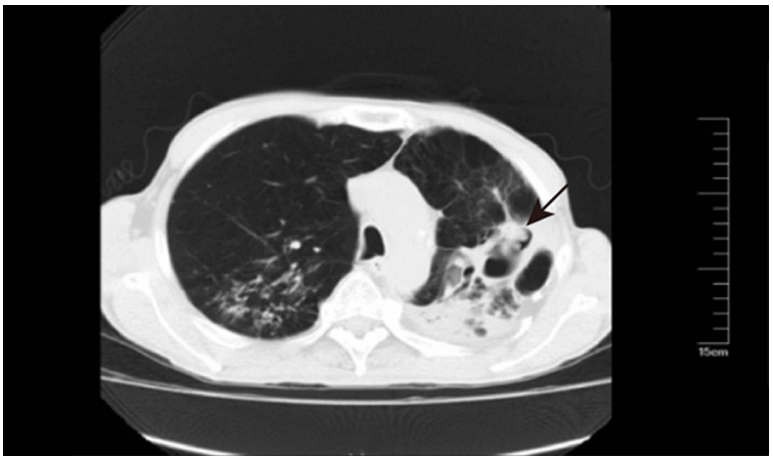

Figure 1. Computed tomography (CT) showing cavity in the left upper lobe with nodular bumps. Black arrow indicated cavity with nodular bumps.

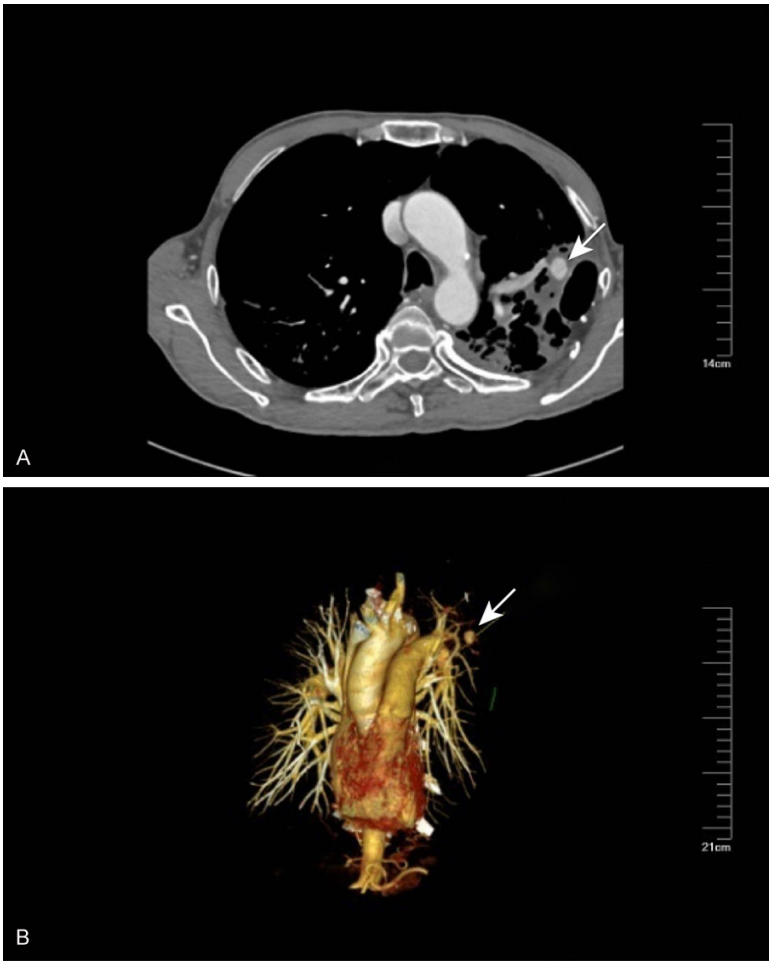

Figure 2. A. High resolution computed tomographical angiography showing peripheral pulmonary artery aneurysm (PPCA) in the cavity of the left upper lobe. B. 3-dependent vascular remodeling revealed a 1.1 cm × 1.7 cm × 1.9 cm (anteroposterior × superoinferior × mediolateral) sized PPCA. White arrow indicated PPCA.

and emphysema, cavity and infection in the left upper lobe with nodular bumps (**Figure 1**). High resolution computed tomographical angiography and 3-dimension vascular remodeling showed a 1.1 cm × 1.7 cm × 1.9 cm (anteroposterior × superoinferior × mediolateral) sized PPCA in the cavity of the left upper lobe (**Figure 2**). The formation of PPCA was less than 6 months after comparison with the previous CT results (**Figure 3**). Pulmonary artery embolization was performed to block the massive hemoptysis. Angiography-guided embolization of the left upper lobe pulmonary artery was done with vascular plug (20 mm) along with fibered coil (3 mm × 40 mm) size (**Figure 4**). After that, massive hemoptysis was still persisted and the patient was offered surgical management. Finally, the patient was treated with left upper lobe sleeve resection to stop the massive hemoptysis. Pathological examination also confirmed inflammatory changes in the left upper lobe (**Figure 5**). The patient stayed in the ICU (Intensive Care Unit) for 2 days. After combined therapy with anti-inflammation, anti-asthmatic, and anti-tussive medications, the patient recovered smoothly and was followed up for more than a year without further recurrence of hemoptysis.

Discussion

PPCA is rare and the exact incidence is unclear [1, 2]. In 1947, Theodoropoulos et al. published a review of PPCA over a period of 100 years and the incidence was report-

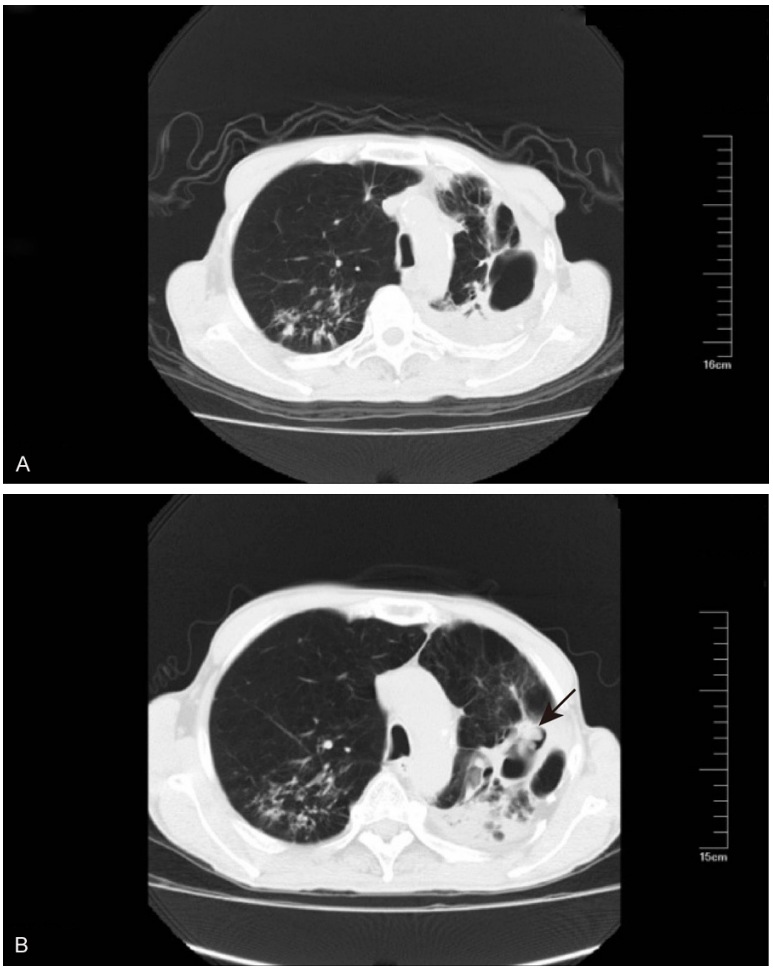

Figure 3. Computed tomography (CT) of this patient at 6 months ago (A) and at present (B). Black arrow indicated PPCA.

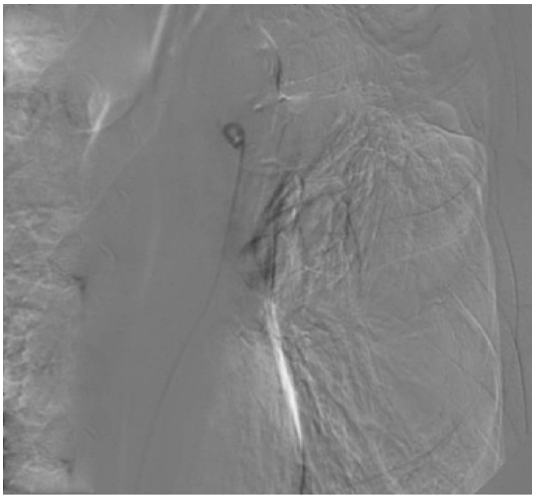

Figure 4. Post-embolization pulmonary angiogram demonstrated occlusion of the aneurysm with vascular plug along with fibered coil.

ed to be 0.0073% [8]. At present, the etiology of PPCA is not completely understood. Approximately one-half of PPCA are related to congenital heart disease accompanied with pulmonary hypertension [9]. PPCA also occurs due to stenosis or absence of leaflets of the pulmonary valve [10]. Other causes may include bacterial endocarditis, syphilis, cystic medial necrosis, tuberculosis, vasculitis, trauma, and hypertension [11, 12]. Behcet's disease is well known to cause multiple aneurysms of the pulmonary and branch arteries. PPCA occurs in approximately 1% of patients with Behcet's disease [13, 14].

PPCA is difficult to diagnose in the clinic, since the manifestations are not specific and may be an incidental finding on chest radiograph. CT and high resolution computed tomographical angiography are the diagnostic modalities of choice for PPCA [15]. There are no definitive therapeutic approaches for this disease because of the paucity of information about its history and the outcome after medical or surgical intervention. The recommended treatment to control massive hemoptysis is percutaneous endovascular and traditional open chest surgical techniques [3, 16]. The therapeutic strategy should be considered for big pulmonary aneurysms regardless of the etiology and underlying disease to prohibit possible rupture, if patients have an acceptably low operative risk.

In our study, we report a 62-year-old male who presented with repeated cough, serious dyspnea, and life-threatening massive hemoptysis. We found a PPCA in the cavity of the left upper lobe with nodular bumps. The formation of PPCA was less than 6 months after comparison with previous CT results. After treatment with anti-inflammation, anti-asthmatic, and an-

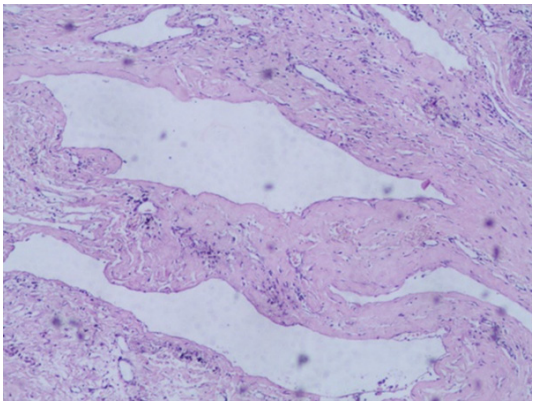

Figure 5. Pathological examination showed inflammatory changes in the left upper lobe (magnification × 100).

ti-tussive medications, pulmonary artery embolization and left upper lobe sleeve resection, the massive hemoptysis was stopped. The patient recovered smoothly and followed for more than a year without further recurrence of hemoptysis. Because pulmonary artery embolization and improved surgery approaches, there are very few deaths about pulmonary artery aneurysm with massive hemoptysis in previous reports. This case of recovery may have the following reasons: (1) Dispose in time, (2) Patient urged to seek operative treatment, (3) Combined treatment with anti-inflammation, anti-asthmatic, and anti-tussive medications, (4) Others. PPCA in the patient was caused by pulmonary tuberculosis and recurrent pulmonary infection. Pathological examination also confirmed inflammatory changes in the left upper lobe. Auerbach in 1939 detected pulmonary aneurysms in 4% of patients dying from tuberculosis [17]. With multi-detector CT, the prevalence of pulmonary aneurysm was detected in a large retrospective series of 189 cases with massive hemoptysis from tuberculosis [17]. The patient had massive hemoptysis after combination PPCA with repeated coughs. After pulmonary artery embolization failed to stop the massive hemoptysis, left upper lobe sleeve resection was suitable to prevent massive hemoptysis.

PPCA is an important cause for massive hemoptysis in patients with tuberculosis. Careful evaluation of the previous contrast CT results may be helpful in suggesting the correctly diagnosis. We recommend that there should be multidisciplinary approaches to achieve optimal patient management in this disease.

Disclosure of conflict of interest

None.

Address correspondence to: Yi Hu, Respiratory Medicine, The Central Hospital of Wuhan, Tongji Medical College, Huazhong University of Science and Technology, 26 Shengli Street, Wuhan 430014, P. R. China. Tel: 0086-27-82211466; E-mail: huyi_pub@126.com

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