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Case Report

Anticoagulant treatment for delayed pulmonary vein stenosis caused by catheter ablation for atrial fibrillation: a long-term follow-up

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Abstract: We present a case with hemoptysis due to delayed pulmonary vein (PV) stenosis 6 months after catheter ablation of atrial fibrillation (AF), which took anticoagulation therapy only and had a good outcome during the long time follow-up. Compared to PV stenosis, the symptom of hemoptysis may lead to the diagnosis of more common diseases. Effectively differential diagnosis is necessary. In conclusion, prevention is of great importance for this new type of iatrogenic disease because there is no satisfied treatment method so far. Anticoagulant therapy may be a new strategy to help fight the disease. The exact effect of warfarin and for how long the anti-coagulation should be remains to be established.

Keywords: Hemoptysis, atrial fibrillation, catheter ablation, PV stenosis, anticoagulant therapy

Introduction

Hemoptysis is a common symptom in the department of respiration, mostly (over 80%) because of bronchiectasis, tuberculosis and lung cancer. Hemoptysis could also arise from other uncommon factors, such as chronic pulmonary aspergillosis, lung abscesses, pulmonary vein (PV) stenosis and cryptogenic causes [1]. PV stenosis, which is usually acquired from catheter ablation of atrial fibrillation (AF), is infrequently considered as a cause of hemoptysis [2, 3]. In this report, we have present a case with hemoptysis due to delayed PV stenosis 6 months after catheter ablation of AF. PV stenosis was usually treated with intervention. In this report, we tried a new treatment strategy and analyzed its effect and long-time outcome. This study was conducted in accordance with the declaration of Helsinki and with the approval of Ethics Committee of Nanjing Medical University. Written informed consent was obtained from all participants.

Case report

A 52-year-old male was admitted to our hospital complaining of left chest pain for a month and cough with intermittent hemoptysis for 2 weeks on Aug.21, 2014. The patient started suffering from left chest pain without cough and radiation to other parts a month ago. A resting 12-lead electrocardiogram showed a T-wave change. There was no abnormity on Chest X-ray. He didn't pay much attention to it until the hemoptysis appeared two weeks ago. The hemoptysis was not massive but intermittently occurred accompanied by a little white phlegm without fever and night sweats. Chest pain kept existing and hemoptysis did not improve. His medical history included hypertension for 7 years and paroxysmal atrial fibrillation (AF) for 2 years. Besides, he took twice radiofrequency ablation-circumferential pulmonary vein isolation (catheter ablation) to AF and had remarkable effect. The first surgery time was Jan.18, 2013 and the last procedure was performed on Jan.14, 2014. His physical examination was unremarkable. Chest CT showed scattered patchy, ground-glass exudative lesions in the top-left lung (**Figure 1Aa**).

The patient was initially diagnosed as pulmonary infection. After a cause (a week) of general anti-infection treatment, the chest pain and

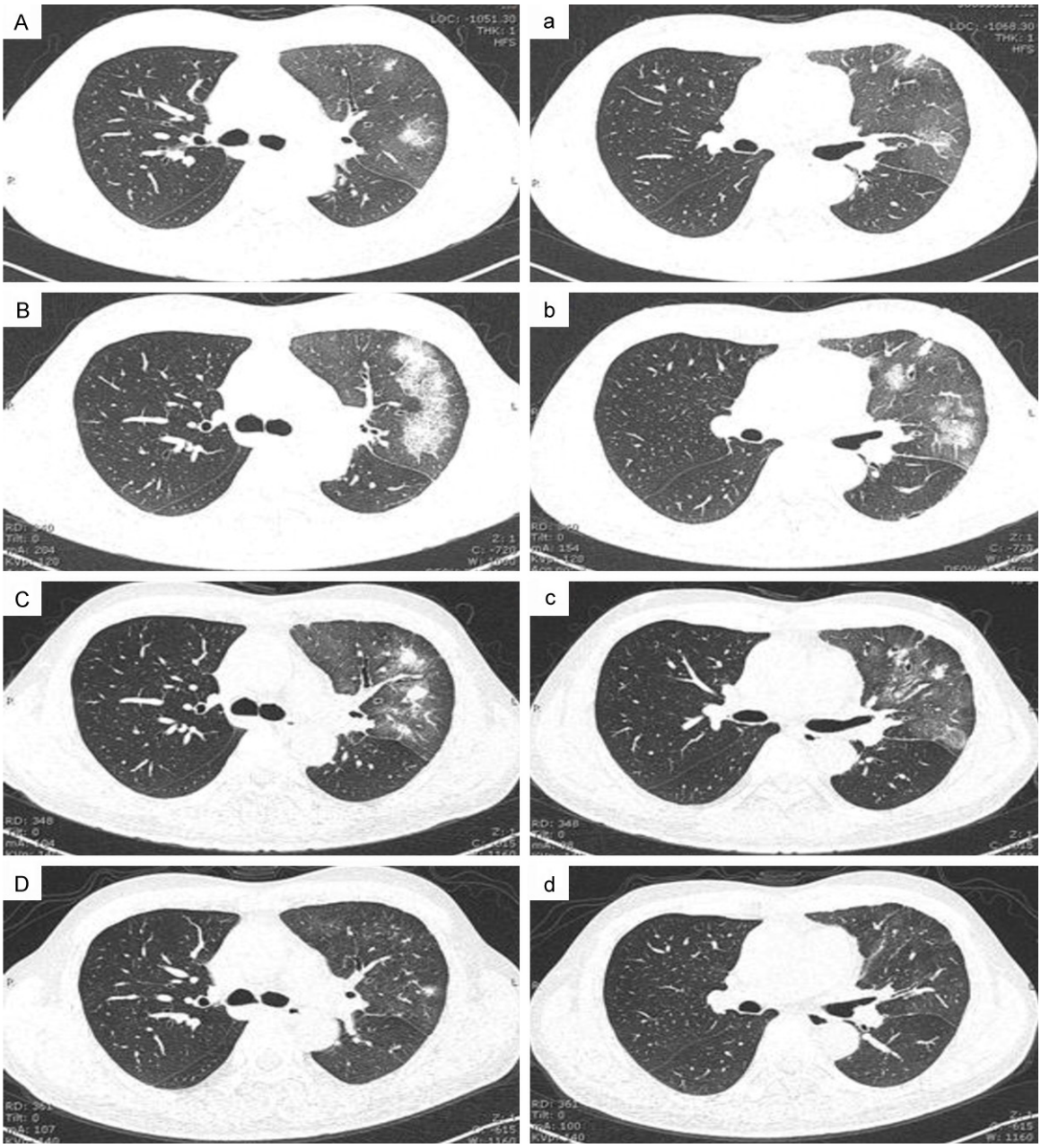

Figure 1. 1-mm thick chest CT image at different stages. Aa. CT showed scattered patchy, ground-glass exudative lesions in the in the upper lobe of the left lung (2014-8-20). Bb. After a course (a week) of general anti-infection treatment increased, repeated chest CT showed enriched exudative lesions and a little pleural effusion in the left lung (2014-8-27). Cc. Repeated CT showed that the exudative lesions in the upper lobe of the left lung were much less than a month ago. The pleural effusion and the scattered patchy exudative lesions in the lower left lobe were almost absorbed completely (2014-9-30). Dd. The latest CT revealed that the exudative lesions in the left lung were almost completely gone (2015-4-10).

hemoptysis were not improved. Used drugs included moxifloxacin, piperacillin, cefoselis, tienam and caspofungin. Repeated chest CT showed increased, enriched exudative lesions and a few pleural effusion in left lung (**Figure 1Bb**). Since Bacterial and fungal culture of sputum and blood samples were negative, anti-infection treatment was ceased. The result of color Doppler echocardiography and transesophageal echocardiography was normal. The result of multi-slice apiral CT pulmonary vein angiography (CTPA) revealed that the left superior PV did not develop and the branches of left inferior PV were mild stenosis (**Figure 2**).

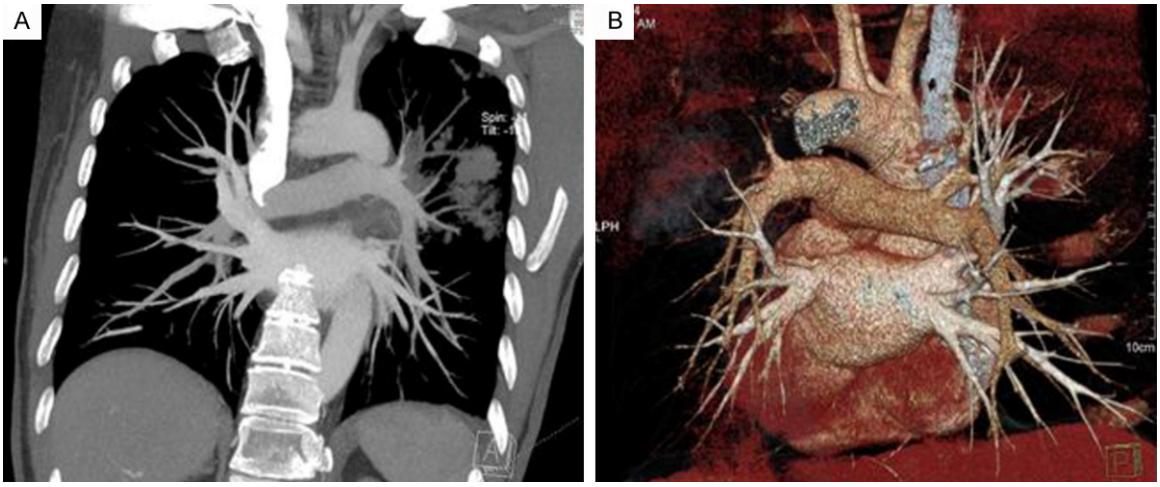

Figure 2. CTPA revealed that the left superior PV did not develop and the branches of left inferior PV were mild stenosis.

Ultimately, the patient was diagnosed as delayed pulmonary vein (PV) stenosis due to catheter ablation of atrial fibrillation (the left superior PV occlusion, the left inferior PV with mild stenosis, and pulmonary congestion in the left superior lung lesions).

The patient was treated by warfarin for anticoagulation instead of any kind of surgery. He kept taking warfarin and has a longtime follow-up. He recovered to normal work 15 days after he left hospital, though he still had a little chest pain and hemoptysis occasionally. A month later, chest pain disappeared. The times and amount of hemoptysis was much less than before. Repeated CT showed that the exudative lesions in the top-left lung were much less than a month ago. The pleural effusion and the scattered patchy exudative lesions in the left lower lobe were almost absorbed (**Figure 1Cc**). The latest CT was 6 months later after discharged from hospital, which revealed that the exudative lesions in the left lung were almost disappeared (**Figure 1Dd**). The symptoms were almost disappeared.

Discussion

In our case, the patient entered into the department of respiration because of hemoptysis and chest pain. Compared to PV stenosis, hemoptysis may lead to the diagnosis of more common diseases. Our first diagnosis was pulmonary infection due to the CT image. It was highly possible to be nosocomial infection because the patient is a doctor of department of hematology. However, the patient was afebrile. And the antibiotics we used basically covered gram-positive and gram-negative bacteria. As the antifungal agents were ineffective, the possibility of fungus infection was also ruled out. The result of T-SPOT.TB was positive, which could not help us rule out the possibility of tuberculosis. But the patient didn't show clinical manifestation of tuberculosis, including fever and night swear. Combining with the negative acid-fast bacilli (AFB) smears and cultures and the normal level of erythrocyte sedimentation rate (ESR), therefore, it was barely possible to be tuberculosis.

The signs of pulmonary alveolar hemorrhage in our patient's chest imaging reminded us of pulmonary vasculitis, which turned out not to be supported by the normal level of ESR, negative of ANCA and glomerular basement membrane antibody. Our patient didn't have the medical history of bronchiectasis. The chest CT also didn't show thickened and dilated bronchus. Another likely cause of hemoptysis is a pulmonary tumor, which was obviously not possible in our case. Besides, cancer cells were not found in sputum cytology test. It's also not convincing to have the diagnosis of pulmonary embolism when the D-dimer and pulmonary arterial pressure were within the normal level.

Considering that the patient had twice catheter ablation for AF and the last time was 6months before this visit, the review of the literature showed that PV stenosis had been described as a life-threatening complication of PV stenosis after successful catheter ablation of AF over the past years. Over the past years, PV stenosis

kept being misdiagnosed due to its unspecific symptoms, such as dyspnea and hemoptysis. Pulmonary vein angiography confirmed the diagnosis of PV stenosis due to catheter ablation for AF in this case. Therefore, a high clinical suspicion and early recognition of PV stenosis should be given to hemoptysis patients with radiofrequency catheter ablation for AF.

Radiofrequency catheter ablation has been proved to be a safe and effective treatment for recurrent, drug-resistant AF. The incidence of PV stenosis has fallen due to the improvement of the procedure and was reported to be 5% to 8% [4]. Symptoms may be developed over the course of 103 ± 100 days of follow-up. Our patient developed hemoptysis and chest pain 6 months after his second procedure. Packer et al. revealed that 52% of the patients with PV stenosis after radiofrequency ablation for AF had undergone a second ablation and only 13% of them developed hemoptysis [5]. The delayed PV stenosis may result from thermal injury to the PVs during the procedure. One likely pathophysiological mechanism is a progressive, irreversible periadventitial inflammation or collagen deposition after extensive RF energy application to canine PVs [6]. PV stenosis could lead to pulmonary congestion, expressed as patchy, exudative lesions in chest CT. Bronchoscopy, which could cause fatal massive hemoptysis, should not be blindly chosen to help diagnose.

Balloon angioplasty and stents implantation are commonly used surgical intervention methods for asymptomatic PV stenosis and the short-term curative effect is remarkable. However, high rate of restenosis (50%) was reported and patients often need a second surgery [7, 8]. Lobectomy was reported to be performed to relieve patients' symptoms when intervention treatment was unsuccessful or not possible [9-11]. In our case, the patient's symptoms didn't getting worse during his hospitalization and pulmonary artery hypertension didn't happen. When taking the surgical trauma into consideration, the patient decided not to take the surgery eventually. Hemoptysis in this case was correlated to secondary thrombosis after PV stenosis. During the formation of collateral circulation, anticoagulants may improve vascular patency and reduce the probability of blood coagulability. Thus, we tried a new treatment strategy. We treated him with anticoagulant therapy only and no intervention was implemented. The patient kept taking warfarin and regular re-examination after he left hospital. His subsequent progress to date has been favorable. We find that early intervention may not be the first choice for symptomatic patients who show the disease mildly and steady-going. Expectant treatment could be a better choice to help fight the disease. It can also avoid the risk of surgery complications and the high restenosis rate effectively. A long time follow-up in this case has revealed its feasibility.

Conclusion

This case highlights a new treatment and diagnosis thinking for hemoptysis patients. A high clinical suspicion and early recognition of PV stenosis should be given to hemoptysis patients with radiofrequency catheter ablation for AF. So far, there is no satisfied treatment method for this complication, which reveals the importance of prevention for this new type of iatrogenic disease. Compared to intervention, simple anticoagulant therapy and a long time follow-up may be more appropriate to some cases. The exact effect of warfarin and for how long the anti-coagulation should be remains to be established.

Disclosure of conflict of interest

None.

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