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Edoxaban treatment for pulmonary arterial thromboembolism associated with azygous vein aneurysm

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Abstract

A 76-year-old woman, diagnosed 5 years previously with an asymptomatic mediastinal bronchogenic cyst, was referred to our department as the mass had grown slightly larger. Contrast-enhanced computed tomography and magnetic resonance imaging scans revealed an azygous vein aneurysm with a pulmonary arterial thromboembolism. The patient was treated with heparin for 5 days, and anticoagulation therapy with edoxaban was continued for 12 months. The thrombus resolved, and the aneurysm remained unchanged. An azygous vein aneurysm is a very rare condition that causes pulmonary arterial thromboembolism. Although surgical resection is indicated for patients with azygous vein aneurysms with a risk of rupture and pulmonary embolism, we chose anticoagulation therapy because of the patient's advanced age. This case suggests that the azygous vein aneurysm is one of the differential diagnoses for a mediastinal mass and that anticoagulation therapy can be the treatment of choice for pulmonary arterial thromboembolism with an azygous vein aneurysm.

Introduction

An azygous vein aneurysm is a very rare cause of a mediastinal mass and sometimes causes pulmonary arterial thromboembolism [1]. The treatment for an azygous vein aneurysm remains controversial, but surgical resection is recommended in cases complicated by rupture, embolism, or obstruction [2]. We present a case of pulmonary arterial thromboembolism due to an azygous vein aneurysm successfully treated with anticoagulation therapy (edoxaban) rather than surgical intervention.

Case Report

The patient was a 76-year-old woman with an asymptomatic 41 mm × 30 mm mediastinal mass. A computed tomography (CT) scan suggested a bronchogenic cyst, and she was observed with regular CT scans for five years. The mass became slightly larger but was asymptomatic. She was referred to our department for examination. A CT with single bolus of contrast and magnetic resonance imaging showed an enhanced anterior mediastinal mass (Fig. 1A, B, white arrow) connected with the azygous arch (Fig. 1A, yellow arrow) and a defect of the A3 right pulmonary artery (Fig. 2A, white arrow). We diagnosed an azygous vein aneurysm with pulmonary arterial thromboembolism.

On the day of admission, venous Doppler sonography of the patient's lower limbs did not reveal deep vein thrombosis in the femoral or popliteal veins, and the D-dimer level was 2.8 mg/L. Laboratory evaluations for thrombophilic parameters revealed normal levels for proteins C and S and anti-cardiolipin antibodies, thus eliminating other causes of hypercoagulability as possible aetiologies of thrombosis.

Anticoagulation therapy with heparin was administered for 5 days and then continued with edoxaban. D-Dimer levels decreased to within the normal range, and there were no side effects.

After 3 months of anti-coagulant therapy with edoxaban, a contrast-enhanced CT scan revealed that the pulmonary arterial thrombosis had disappeared, and the azygous vein aneurysm did not increase in size (Fig. 2B). After 12 months of edoxaban therapy, D-dimer remained

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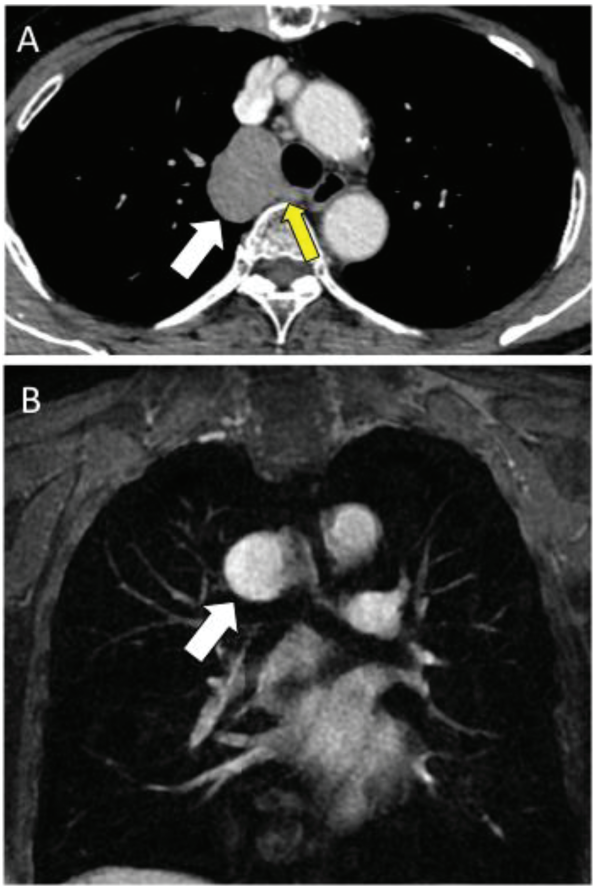

Figure 1. Computed tomography with single bolus of contrast (A) and magnetic resonance imaging (B) showed an azygous vein aneurysm (white arrow) connected with the azygous arch (yellow arrow).

within normal levels, and there was no recurrence of pulmonary arterial embolism.

Discussion

We present a case of an azygous vein aneurysm with pulmonary arterial thromboembolism that was considered to be a bronchogenic cyst for five years. An azygous vein aneurysm is a rare condition, and most cases are asymptomatic. Three main causes of an azygous vein aneurysm are reported [3]: (1) pressure and volume overload, such as congestive cardiac failure and portal hypertension; (2) traumatic pseudoaneurysm; and (3) idiopathic aneurysm. In our case, according to medical history, physical examination, and imaging findings, the cause is considered idiopathic.

In few cases, an azygous vein aneurysm has been a comorbidity with pulmonary arterial thromboembolism due to the thrombus in the aneurysm. We eliminated other possible causes of thrombosis and diagnosed pulmonary arterial thromboembolism due to the azygous vein aneurysm.

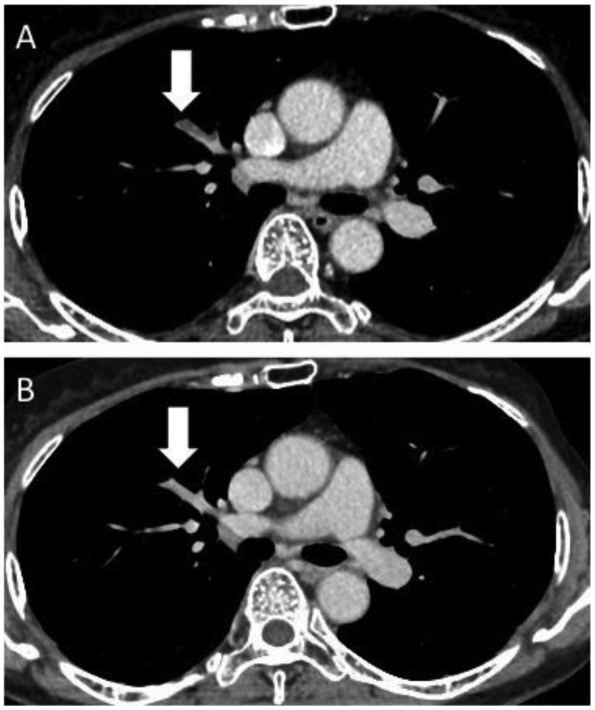

Figure 2. (A) A contrast-enhanced computed tomography scan revealed a defect of the A3 right pulmonary artery (white arrow). (B) After 3 months of anticoagulant therapy with edoxaban, the pulmonary arterial thrombosis had disappeared (white arrow).

In our case, in the unenhanced CT scan, the azygous vein aneurysm mimicked the appearance of a mediastinal mass. The feature of a dynamic CT scan of an azygous vein aneurysm is mild enhancement in the early arterial phase and delayed enhancement [4]. A dynamic CT scan should be performed in order to diagnose an azygous vein aneurysm (which may cause pulmonary arterial thromboembolism) in patients with a mediastinal mass.

Although the treatment of azygous vein aneurysms remains controversial, surgical resection is recommended in cases complicated by embolism. In our case, we chose treatment with anticoagulation therapy (edoxaban) because of the patient's advanced age. Edoxaban is an oral non-vitamin K antagonist anticoagulant and is known to be effective for pulmonary thromboembolism [5]. To the best of our knowledge, the efficacy of edoxaban for pulmonary thromboembolism due to an azygous vein aneurysm has not been reported. There was no recurrence of pulmonary thromboembolism after 12 months of edoxaban maintenance therapy.

We learned two lessons in this case: (1) an azygous vein aneurysm, which may cause pulmonary thromboembolism, is an important differential diagnosis for a mediastinal mass; and (2) edoxaban anticoagulant therapy should be considered for patients with this condition.

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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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