American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2018; 19: 1301-1305
DOI: 10.12659/AJCR.910982

Received: 2018.05.06
Accepted: 2018.08.09
Published: 2018.11.01

An Autopsy Case of Paradoxical Pulmonary Thromboembolism by Arteriovenous Fistula from Right Common Iliac Artery to Common Iliac Vein: A Rare Cause of Pulmonary Embolism

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Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Conflict of interest: None declared

Patient: Male, 74
Final Diagnosis: Paradoxical pulmonary thromboembolism
Symptoms: Dyspnea
Medication: —
Clinical Procedure: Autopsy
Specialty: Pulmonology

Objective: Rare co-existance of disease or pathology

Background: Pulmonary thromboembolism (PTE) sometimes leads to a shock state and sudden death due to acute massive pulmonary arterial thrombosis. The origins of pulmonary arterial thrombi are varied, but most arise from deep vein thrombosis. We herein presented a very rare cause of PTE due to paradoxical embolism caused by arteriovenous fistula from common iliac artery to common iliac vein.

Case Report: A 74-year-old man was admitted because of increasing dyspnea on exertion. The diagnosis of idiopathic pulmonary fibrosis was made and corticosteroid therapy was started. On the 5th hospital day, the patient suddenly developed cardiopulmonary arrest and died despite cardiopulmonary resuscitation. An autopsy revealed that the left main pulmonary artery was occluded by a massive but organized thrombus that was similar to an arterial thrombus in the right common iliac artery. On histopathological examination, the emboli in the pulmonary artery demonstrated characteristics similar to thrombus in the common iliac artery. This suggested that the emboli had passed through the arteriovenous fistula from the right common iliac artery to the common iliac vein.

Conclusions: This is a very rare cause of fatal paradoxical pulmonary thromboembolism. Paradoxical emboli passing through arteriovenous fistula from the right common iliac artery to the common iliac vein are a rare cause of PTE.

MeSH Keywords: Arteriovenous Fistula • Iliac Artery • Pulmonary Embolism

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/910982

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Background

Pulmonary thromboembolism (PTE) sometimes leads to a shock state and sudden death due to acute massive pulmonary arterial thrombosis and obstruction leading to acute right heart failure. Deep vein thrombosis (DVT) complicated by PTE has various etiologies, including congenital or acquired coagulation-fibrinolytic disorders, blood stagnation in major vessels, and injuries of major venous vascular walls [1]. We recently had a patient who suddenly died from PTE. An autopsy revealed a thrombus in the pulmonary artery derived from an arteriovenous fistula, with characteristics similar to those in the common iliac artery. We report this case as a very rare cause of PTE.

Case Report

A 74-year-old man was admitted to our institute because of dyspnea and cough. He reported loss of appetite and weight, with dyspnea on exertion and cough for 2 months prior to admission. Pulmonary fibrosis was noted on a chest X-ray 3 years prior to admission. He had a 100-pack-year smoking history and essential hypertension. On admission, his vital signs were as follows: body temperature, 37.6°C; pulse rate, 65 beats per min; blood pressure, 108/56 mmHg; respiratory rate, 20 breaths per min; and arterial oxygen saturation of 92% when inhaling oxygen from a mask with a 24% fraction of inspired oxygen (FiO_2). Physical examination showed no anemia or jaundice, no jugular venous dilatation, and no leg edema. Auscultation revealed normal heart sounds and inspiratory fine crackles at the bilateral lower lung fields. There was no peripheral edema, abdominal bruit, or other lower-extremity abnormality. As shown in Figure 1, chest X-ray and computed tomography (CT) demonstrated a chronic fibrosing interstitial pneumonia pattern that gradually progressed within 1 week. Laboratory tests on admission revealed mild anemia (Hb 9.7 g/dL) and slight elevation of LDH and C-reactive protein, and moderately increased serum KL-6 and surfactant protein D, suggesting exacerbation of interstitial lung disease (Table 1). Unfortunately, D-dimer or other coagulation and fibrinolytic parameters were not measured. Electrocardiogram revealed no significant ST-T change and no severe right ventricular overload, as shown in Figure 2. Therefore, on admission we suspected that the patient's dyspnea was due to respiratory impairment caused by interstitial pneumonia, and cardiac factors like right heart failure might have had a relatively low contribution.

The diagnosis of acute exacerbation of idiopathic interstitial pneumonia was made and corticosteroids were started: 20 mg/kg/day of methylprednisolone for 3 days, followed by 1 mg/kg/day of prednisolone. Low-grade fever and dyspnea on exertion were slightly improved by the 3rd hospital day. On the 4th hospital day, however, the patient had cardiopulmonary arrest on the ward, and he died despite cardiopulmonary resuscitation.

An autopsy revealed white emboli, which were massive but thin and organized, in the left main pulmonary trunk (Figure 2). Furthermore, microscopic vascular embolisms of pulmonary artery branches of both right upper and lower lobes, as well as left upper and lower lobes, appeared. These microscopic embolisms were thought to be fresh. The right common iliac artery was

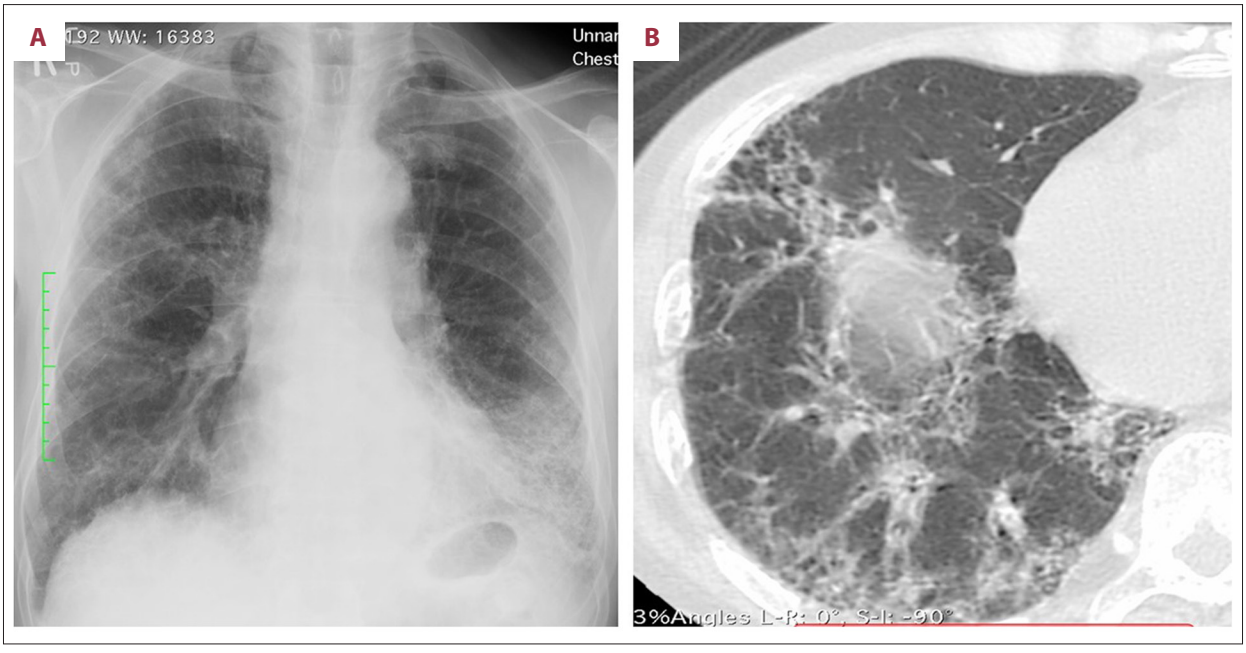

Figure 1. Chest X-ray (**A**) and CT (**B**) demonstrated a chronic fibrosing interstitial pneumonia pattern that was gradually progressive.

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Table 1. Blood laboratory tests on admission.

Blood cell count			**Serum chemistry**		
WBC	6600	/μL	Albumin	3.1	g/dL
Hb	9.7	g/dL	Cre.	0.84	mg/dL
plt.	134 000	/μL	BUN	14	mg/dL
Coagulation			LDH	351	IU/L
PT	52	%	Na	136	mEq/L
PT-INR	1.28		K	3.7	mEq/L
APTT	36	s			
			KL-6	1510	U/mL
Arterial blood gas (room air)			SP-D	144	ng/mL
pH	7.44		CRP	5.9	mg/dL
PaO2	72.9	mmHg			
PaCO2	30.3	mmHg			
$HCO3^-$	20.3	mEq/L			

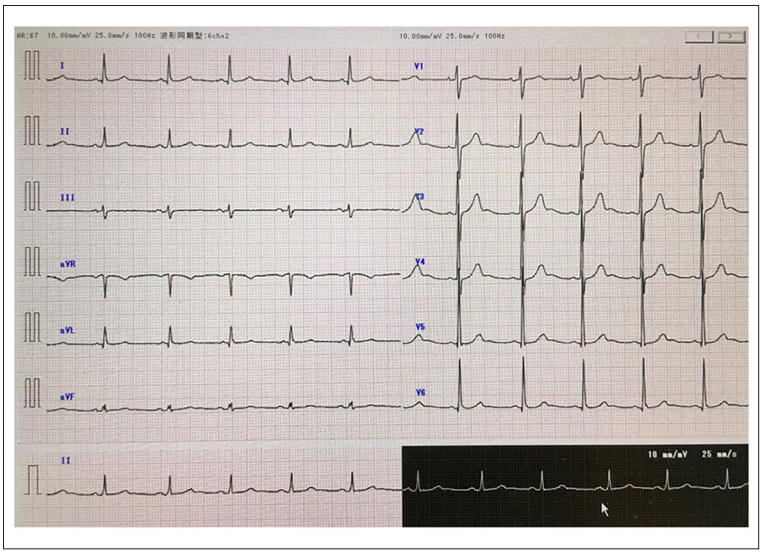

Figure 2. Electrocardiogram showed normal sinus rhythm, left ventricular hypertrophy, and no significant right ventricular overload.

found to have an aneurysm that extended to the right common iliac vein to create an arteriovenous fistula (Figure 3). A significant thrombus, which was thought to have originated from a major vein, could not be found in the autopsy. A thrombus found in the right common iliac artery was macroscopically and histologically similar to an embolus in the left pulmonary trunk, with fine crystalloid nodules scattered throughout the thrombus (Figure 4). Although the origin of this crystalloid nodular lesion was not determined, it was suspected to be calcification. We concluded that the fatal PTE in this case was caused by paradoxical emboli that had passed through an arteriovenous fistula formed by penetration from the right common iliac artery to the vein.

Discussion

We reviewed our management of this case regarding the prediction and prophylaxis of PTE. An examination for a major abdominal vascular abnormality at the first patient visit or on admission, such as ultrasonography or CT with contrast, might have been useful. However, no pulsating mass or audible vascular bruit was found, and rescue therapy for acute exacerbation of idiopathic interstitial pneumonia with corticosteroids had priority. It is conceivable that the dyspnea on exertion and hypoxia in this patient were caused not only by interstitial pneumonia, but also by PTE. In clinical practice, however, it may be difficult to diagnose pulmonary arterial embolization complicated by an arteriovenous fistula, especially when other

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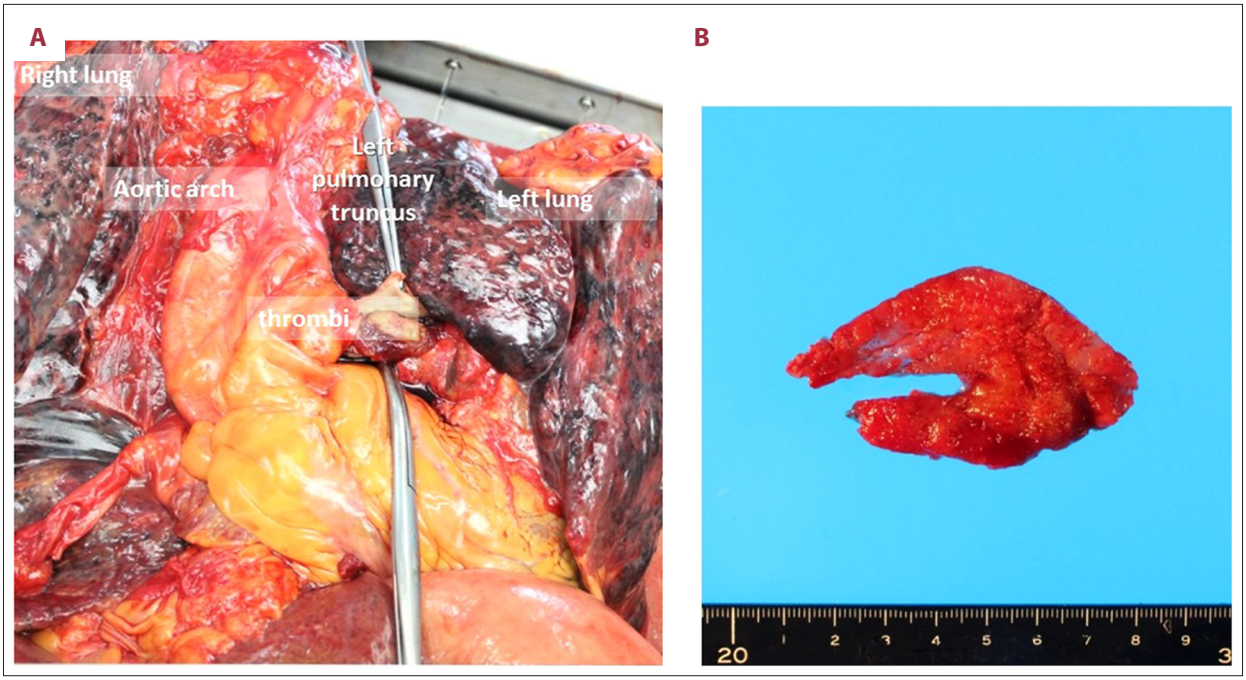

Figure 3. (**A**) Shows the gross appearance of the left main pulmonary trunk at autopsy; (**B**) shows an organized thrombus in the left pulmonary artery.

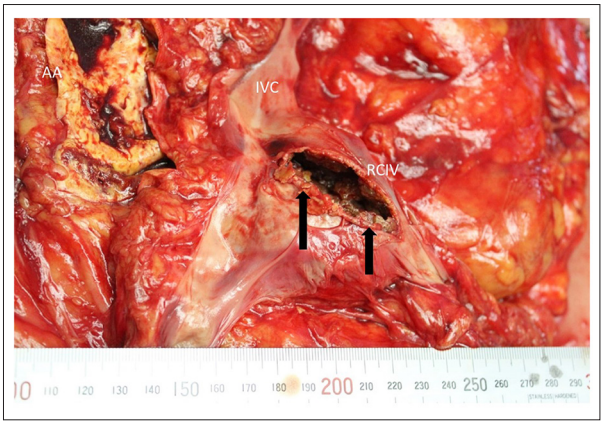

Figure 4. The right common iliac artery formed an aneurysm that penetrated the right common iliac vein to create an arteriovenous fistula (arrows).

pulmonary diseases are apparent on routine physical examination and laboratory or radiological tests.

Acute PTE in association with DVT is considered to have 3 components: i) a congenital or acquired coagulation or fibrinolytic abnormality, ii) a disturbance of blood flow in the major veins, and iii) an injury of the vascular wall of a major vein [1,2]. In this case, we attributed the development of PTE to paradoxical embolization associated with an arteriovenous fistula from the

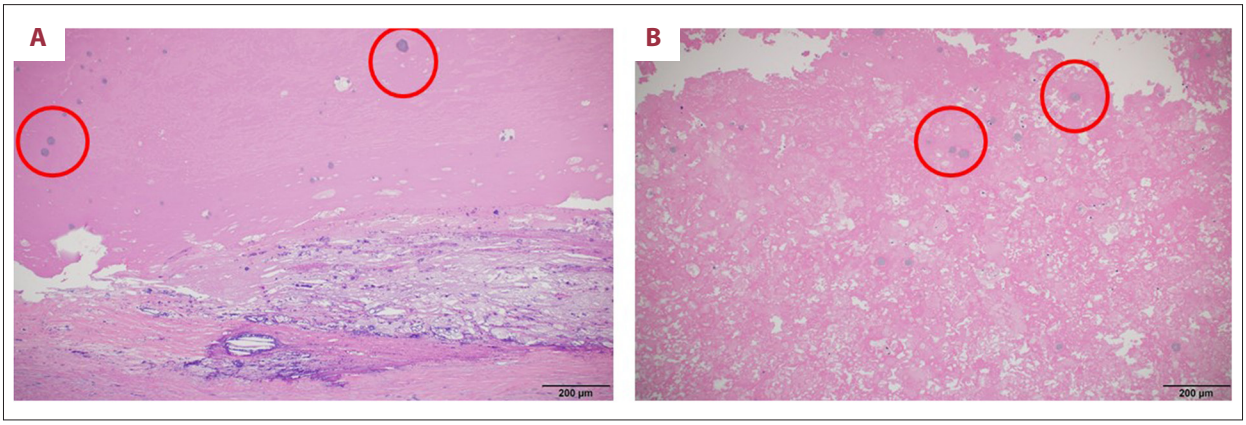

Figure 5. Thrombus found in the right common iliac artery (**A**) was histologically similar to embolus in the left pulmonary trunk (**B**), as fine crystalloid nodules were scattered throughout the thrombus (circles).

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right common iliac artery to the left common iliac vein, which is considered a very rare cause of PTE.

Arteriovenous fistulas are present in 0.2–1.3% of patients with an abdominal aortic aneurysm (AAA) and are found in 3–4% of patients with a ruptured AAA [3]. The inferior vena cava is the most common site of an arteriovenous fistula associated with an AAA, and the common iliac vein is the second most common site [4,5]. Reports of complications from major venous vessel embolization, in association with an arteriovenous fistula of the common iliac vein, are rare [4,6,7]. Furthermore, case reports describing complications of PTE have been rare [8–10], although a recent review [11] described not only a case report but also diagnosis by helical CT [12] and management with endovascular intervention [13,14]. Bridger reported an 83-year-old woman with a paradoxical PTE and thrombosis in a ruptured AAA involving the inferior vena cava that was diagnosed at autopsy [15]. However, a paradoxical PTE complicated by an arteriovenous fistula from the common iliac artery to the iliac vein, as in our case, has not been reported.

The embolus found in the left pulmonary trunk was histologically similar to the thrombus in the right common iliac artery, as fine crystalloid nodules were scattered throughout the thrombus and the embolus (Figure 5). Although the origin of these crystalloid nodules was not determined, they may have been due to calcifications that are often seen in atherosclerosis and arterial plaque. Similar reports have rarely described thrombi in histological detail. In this case, histology suggested that PTE was due to pulmonary arterial paradoxical embolization, with a thrombus passing from the right common iliac artery to the vein through an arteriovenous fistula, which had been formed by penetration from the iliac artery.

Conclusions

In this report, we have described an autopsy case of PTE due to paradoxical embolization with a thrombus in the common iliac vein. The thrombus had passed from the right common iliac artery through an arteriovenous fistula formed by penetration from the iliac artery to the vein, leading to sudden death. While it is very rare, clinicians should be aware of the possibility of paradoxical embolization, as in this case.

Acknowledgements

We especially thank the patient and his family, assistant professor, Dr. Yoko Ito, all members of the nursing and care team, and technicians of the Department of Pathology in our institute.

Conflict of interest

None.

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