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## Case Report

# A Rare Case of Peripheral Pulmonary Artery Aneurysm and Cavitating Pneumonia in a Patient with COVID-19 Managed with an Endovascular Method <sup>☆</sup>

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### ABSTRACT

Peripheral pulmonary artery aneurysm (PAA), being a rare condition, is considered extremely rare following coronavirus disease 2019 (COVID-19). We present a 58-year-old male who presented with fever, malaise, and dry cough. SARS-CoV-RNA transcription-mediated amplification test was positive for the patient. After 2 days, he developed hemoptysis and back pain, and a CT scan revealed a pulmonary aneurysm, evidence of alveolar hemorrhage, and Necrotizing pneumonia. He was scheduled for pulmonary artery angiography. The angiography confirmed a fusiform aneurysm and partial coiling of the aneurysmal sac, and indoor and backdoor embolization was performed. In the follow-up, a CT scan showed complete thrombosis of the aneurysmal sac, and the patient was free of symptoms. Peripheral PAAs can show a variety of symptoms. They can even be asymptomatic. The infectious pathologies of this condition are less common than the other. COVID-19 is an extremely rare pathology. To the best of our knowledge, this is the first case of necrotizing pneumonia and peripheral PPA in an adult. Moreover, it was followed by COVID-19. A vital takeaway note for physicians is to consider PAAs as a complication when treating COVID-19 patients who don't show signs of improvement or even show signs of exacerbation.

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## Introduction

Pulmonary artery aneurysm (PAA) is a rare entity constituting about 1% of all intrathoracic aneurysms [1]. The peripheral PAA is even less common (11%) [2]. Peripheral PAA following novel coronavirus disease 2019 (COVID-19) is extremely rare [3]. Here we are presenting a case of necrotizing pneumonia (NP) and peripheral PAA following COVID-19 managed with an endovascular method.

## Case report

A previously healthy unvaccinated 58-year-old male presented to our hospital with 3-day history of fever, generalized malaise and dry cough during the Delta variant outbreak of COVID-19 in our region. The SARS-CoV-RNA transcription mediated amplification test was positive. Two days later, the patient developed severe back pain and an episode of non-massive hemoptysis. An enhanced chest computed topographic (CT) Scan revealed several patchy ground glass density area in periphery and peribronchovascular interstitium of both lungs (Fig. 1b). An oval pulmonary aneurysm was also detected in medial aspect of left lower lobe adjacent to a 40\*14-mm fluid filled cavity (Fig. 1a). Moreover, the evidences of alveolar hemorrhage were seen in parenchyma of left lower lobe. Necrotizing pneumonia was diagnosed and the patient was scheduled for pulmonary artery angiography.

After insertion of a 6F vascular sheath in the right femoral vein, the right pulmonary artery (PA) was catheterized with a 5-F multipurpose (MP) catheter. The angiography confirmed a fusiform aneurysm (13 × 11 mm) arising from the descending branch of the left pulmonary artery Fig. 1d. Afterward, a 2.8F microcatheter (Embocath, Merit Medical) was advanced coaxially into the MP catheter over a 0.014 inch balanced middle weight (BMW) microwire (Abbot Vascular, Santa Clara, CA) to catheterize the feeding artery and the aneurysmal sac.

To exclude the PAA partial coiling of the aneurysmal sac and indoor and backdoor embolization with glue was applied. First, a detachable three-dimensional coil (12 mm × 30 cm, GCD-18 3D Stryker, MI) was inserted for partial coiling of the sac (Fig. 1e). Afterward, the microcatheter was advanced meticulously to the feeding and draining braches, respectively. The most proximal part of the draining branch, the most distal part of the feeding artery and the adjacent parts of the aneurysmal sac were entirely occluded with high concentration n-Butyl Cyanoacrylate (NBCA) (Glubran 2) diluted in Ethiodized oil (Lipiodol, Ultra fluid, Guerbert, France; 70%-30%) (Fig. 1f). After a few minutes, the confirmation angiography was done. The aneurysmal sac was completely excluded from the pulmonary artery vasculature in completion angiography.

In the following day hemoptysis was stopped. The follow up CT scan revealed complete thrombosis of the aneurysmal sac. The patient was discharged with stable condition the day after PA embolization. After 2 months the patient was symptom free.

## Discussion

Although a few cases of pulmonary artery have been reported in COVID-19 patients with acute respiratory distress syndrome on extracorporeal membrane oxygenation [4], mucormycosis [5] and tuberculosis [6], to best of our knowledge it is the first case of NP and peripheral PPA in adult. The first pediatric case was a 13-year-old boy [3].

PAA is a rare pathology found in 1 of 14,000 autopsies [7]. The left lung is a more common location for peripheral PAAs [7]. The dissection and rupture of PAA are catastrophic and can lead to death in 50%-100% of case [8,9]. Therefore early diagnosis is critical.

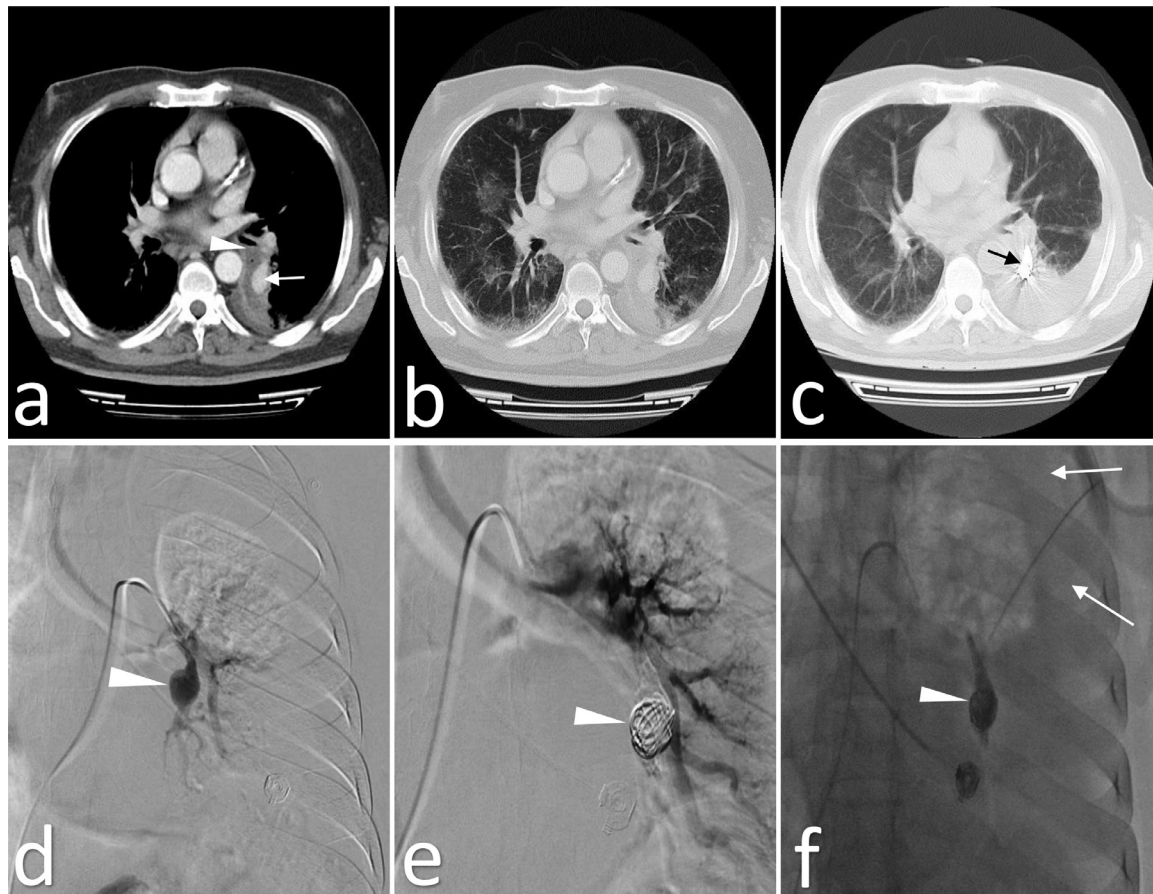
Peripheral PAAs may present with a wide spectrum of sign and symptoms. The patients can be asymptomatic [10]. The pressure effect on surrounding structures may result in non-specific symptoms [10] such as cough, cyanosis, bronchiectasis, pneumonia, dyspnea [11–17], palpitation, hoarseness, and syncopal attacks [18]. Hemoptysis, massive or recurrent small volume, is another presentation [19]. Dramatically, sudden cardiac death may be the first presentation [20].

Congenital cardiac pathologies are the most common cause of PAA [18,21]. Most of these pathologies result in central PAAs; however, the peripheral PAAs are usually detected in connective tissue disorders. About 8% of the patients with Eisenmenger also suffer from peripheral PAAs [21].

The infectious pathologies are less common causes of PAAs involving both main and peripheral PAs. The most common infectious causes of peripheral PAAs are atypical mycobacteria and Tuberculosis especially after reactivation in upper lobes [22,23], pyogenic bacterial infections (Staphylococcus species, Streptococcus, Klebsiella, Actinomyces) [23,24], septic embolism related to bacterial endocarditis [18,25], fungal pneumonia (Aspergillosis, Candida albicans, coccidiomycosis, mucormycosis) [23,24] and parasitic pathogens (schistosomiasis) [21].

Peripheral PAA is more common in patients with vasculitis, autoimmune disorders and idiopathic aneurysms. Multiple bilateral PAAs especially in right lower lobe and main pulmonary arteries is the most common presentation of Behcet Disease [23]. Similarly, Hughes-stovin syndrome is associated with peripheral PAA. [23]. Giant cell arthritis [26] is also a rare cause of peripheral PAA. At last, about two-thirds idiopathic PAAs are intraparenchymal [21].

The location of PAAs in patients with malignant pulmonary masses, trauma and iatrogenic insults has been reported exclusively in peripheral pulmonary vasculature. Squamous cell carcinoma is the most common subtype of pulmonary malignancy associated with PAAs [27]. Metastatic masses [28], primary pulmonary leiomyosarcoma [29] and malignant fibrous histiocytoma [30] are the other masses related to PAAs. Swan-Ganz catcher insertion is the leading iatrogenic cause of peripheral PPA. Pneumonectomy [31], cardiac catheterization [32] and embolising acrylate from porto-systemic shunt after sclerotherapy of an esophageal varices [33], lung biopsy [18] and chest tubes [18] are the less common causes of PAAs. Some patients with peripheral PPA have also been victims of blunt trauma, gunshot, and rib fracture [34].



**Fig. 1 – Chest CT scan Digital subtraction angiography of Pulmonary Artery; a: Pre-embolization Contrast enhanced chest CT scan in mediastinal window revealing a fusiform pulmonary artery aneurysm (arrow) adjacent to a cavity with air-fluid level (arrowhead). b: Pre-embolization Contrast enhanced chest CT scan in lung window revealing several patchy ground glass density area in periphery and peri-bronchovascular interstitium of both lung typical for COVID-19. c: Post-embolization unenhanced chest CT scan in lung window one month after embolization revealing relative resolution of ground glass density. Note high complete embolization of aneurysm (arrow). d: Digital subtraction angiography of the descending branch of left pulmonary artery revealing a fusiform aneurysm (arrowhead). e: Digital subtraction angiography of left pulmonary artery after embolization demonstrating complete exclusion of the aneurysmal sac (arrowhead). f: X-Ray of left lung revealing complete occlusion of the aneurysm (arrowhead). Note pleural effusion (arrows).**

There is no consensus on management of PPA. Some authors propose the watchful waiting for stable asymptomatic PAAs without PAH [18,20,35] especially if they are idiopathic [36]. Treatment of the underlying cause is another logical step in conservative management. However, close monitoring and radiographic follow-up are mandatory [21,37]. Although some medications are available to control the PAH, even normalization of the PA pressure does not avoid further enlargement of PA. In fact, the patients with PAH are the major candidate for nonconservative management [14,18].

Nonconservative management, surgical or interventional, are the mainstay of treatment in symptomatic patients and in patients with PAH [21]. However, surgery carries a high risk of complication and mortality especially in patients with PAH. Endovascular intervention is a good choice when conservative management fails. Transarterial embolization of the aneurysm is the primary purpose of endovascular treatment. Several embolizing agents have been applied such as coil

[25,38,39], vascular plugs [10,40], glue [41] and detachable silicon balloon [42]. Stent graft [43,44] has been also inserted to exclude the aneurysm from pulmonary vasculature. In the event of a bronchopulmonary shunt, embolization of pulmonary branch is not sufficient and bronchial and systemic feeding artery embolization is suggested [8].

In conclusion, the peripheral PAA is a rare entity with several infections, congenital, inflammatory, autoimmune, malignant, and trauma-related etiologies. Severe COVID-19 infection with necrotizing pneumonia seems to be another etiology. Endovascular management is an effective method for management of these patients.

#### Patient consent statement

Consent for publication was obtained for every individual person's data included in the study.

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