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Case Report

Pseudo-vanishing lung syndrome in a patient with tricuspid valve bacterial endocarditis

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ABSTRACT

Infective endocarditis is a major cause of morbidity and mortality among individuals with opioid use disorder who use injection drugs. It is frequently associated with tricuspid valve endocarditis and *Staphylococcus aureus* bacteremia, with secondary pulmonary septic emboli. Herein, we report a unique case of pulmonary cavitation injury following pulmonary septic emboli in the setting of tricuspid valve endocarditis in an injection drug user with opioid use disorder. The pattern of cavitary lung injury mimics radiographically indistinguishable features from vanishing lung syndrome during the most advanced stage of her illness.

<**Learning objective:** This manuscript aims to highlight a new complication of bacterial endocarditis secondary to septic emboli showered from the infected tricuspid valve. This complication, which resembles a pulmonary disease by the name of vanishing lung syndrome, is characterized by extensive pneumatoceles that give the appearance of vanishing lung on chest radiography.>

Introduction

Infective endocarditis (IE) is a major cause of morbidity and mortality among individuals with opioid use disorder (OUD) who inject drugs [1]. IE among injection drug users (IDU) with OUD are often right-sided, mostly involving the tricuspid valve, and usually associated with complications such as pulmonary septic emboli [1]. *Staphylococcus aureus* is one of the most common causes of IE among intravenous (IV) drug users leading to pulmonary septic emboli [2]. Besides, *S. aureus* lung infections are commonly associated with a serious complication known as pneumatoceles [2]. Pulmonary pneumatoceles are thin-walled, gas-filled cysts that develop within the lung parenchyma. They can be single emphysematous lesions, but are more often multiple. The exact pathogenesis of pneumatoceles is uncertain in most cases, but believed to be due to the formation of a mucus flap acting as a check-valve in small draining bronchioles secondary to inflammation and irritation of their walls [3]. Consequently, air is trapped in the bronchial wall, forming large air-filled sacs under tension. These appear as hyper-lucent areas on chest radiographs.

Similarly, radiographically hyper-lucent lungs can be seen in patients with emphysematous disease, a common respiratory illness seen among hospitalized patients. In particular, an entity called idiopathic giant bullous emphysema or vanishing lung syndrome (VLS), is a rare emphysematous disease first described in 1937 by Burke [4], and characterized by specific radiological criteria including the presence of giant emphysematous bullae in one or both of the upper lobes of the lung, occupying at least one-third of the hemithorax, and compressing the surrounding normal lung tissue [5]. There are several reported cases of VLS in the literature, particularly occurring among young males, and risk factors include smoking, marijuana abuse, alpha-1-antitrypsin deficiency, and systemic diseases such as Marfan's syndrome or Ehlors–Danlos syndrome [6,7]. On the contrary, mature pneumatoceles appear as thin-walled cystic spaces within the lung parenchyma, containing air, and usually occur on the fifth to sixth day of hospitalization. Nonetheless, radiographical differentiation of large pneumatoceles that compress normal surrounding lung and emphysematous bullae may be indistinguishable at times.

The association of extensive, large pulmonary pneumatoceles with IE has never been reported in the literature. Therefore, we

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report a case of a 20-year-old female with a history of OUD presenting with tricuspid endocarditis, and complicated by pulmonary septic emboli and pneumatoceles resembling VLS during her hospital stay.

Case *report*

A 20-year-old woman with OUD and IDU was admitted with generalized pain, cough, nausea, vomiting, and shortness of breath. The patient was a smoker and had used intravenous heroin, with a past medical history that was significant for major depressive disorder. Physical examination was only positive for tachycardia and desaturation (oxygen saturation of 64%), with bilateral basal rales, and abdominal tenderness. Laboratory investigations revealed leukocytosis, anemia, and thrombocytopenia, with elevated C-reactive protein and blood-urea-nitrogen/creatinine ratio (BUN/Cr = 37). Blood cultures revealed bacteremia with methicillin-sensitive *S. aureus* (MSSA), and thus the patient was given oxacillin but was switched to vancomycin due to drug-related rash. Transesophageal echocardiogram showed tricuspid valve vegetations and mild tricuspid regurgitation, and thus the patient was diagnosed with tricuspid valve IE. Work-up with computed tomography (CT) scan revealed lesions in the lungs diagnosed as septic emboli (Fig. 1). She was then intubated for respiratory failure and developed acute kidney injury requiring continuous veno-venous hemodialysis. One week after presentation, repeat CT scan showed marked progression of her lung condition with development of cavitations of the septic emboli and bilateral pleural effusions (Supplementary Fig. S1a and b). Pleural fluid was exudative, and cultures grew MSSA. Two weeks following admission, the patient had undergone tricuspid valve replacement and decortication of the right lung, which confirmed the presence of large tricuspid vegetations destroying the entire valve. Pathology of the explanted valve showed inflammation and Gram-positive organisms and the valve cultures grew MSSA. Sixty days following admission, the patient had been clinically stable with persistent dyspnea, on 3 l of oxygen, and undergoing respiratory rehabilitation therapy. However, X-ray and CT scan images showed a pattern of lung injury described as large air spaces, which were suspected to be either bilateral pneumatoceles or multi-septated pneumothoraces (Fig. 2a and b), resembling a pattern consistent with VLS, severely compressing adjacent lung. Alpha 1 antitrypsin levels were measured but were within normal range in this patient. The patient also developed neurologic complications with focal seizures. The cardiothoracic surgeons did not recommend any surgical intervention, and the patient completed 87 days of antibiotic therapy. Over the course of 4 months following her hospital discharge, the patient's radiologic lung abnormality as well as her physical functions improved remarkably, and follow-up CT scans showed near-complete resolution of her lung disease that was first seen during her acute medical illness (Fig. 3).

At the time of discharge, chest X-ray was done and showed small hydropneumothorax and marked improvement of cystic lesions (Fig. 3a). Follow up CT 1 month after discharge also showed near complete resolution except for a few, small resolving, presumed pneumatoceles and residual linear/curvilinear scars (Fig. 3b). Unfortunately, the patient died of drug overdose at her home 2 months after discharge (210 days following her valve replacement surgery) (Fig. 4).

Discussion

Infections are the leading cause of hospitalization (31%) among individuals with OUD, and IE is one major infection contributing to the morbidly and mortality of this patient population [1]. These patients usually suffer from right-sided – particularly tricuspid – IE complicated by multiple septic pulmonary embolisms. In fact, and seen in our patient, *S. aureus* bacteremia, and septic pulmonary emboli are considered a triad pathognomonic for tricuspid valve endocarditis [2]. Pulmonary complications of septic emboli secondary to IE in patients with IDU and OUD include pneumonia, lung abscess, pleural effusion, empyema, fatal pulmonary hemorrhage caused by rupture of mycotic aneurysms of the pulmonary

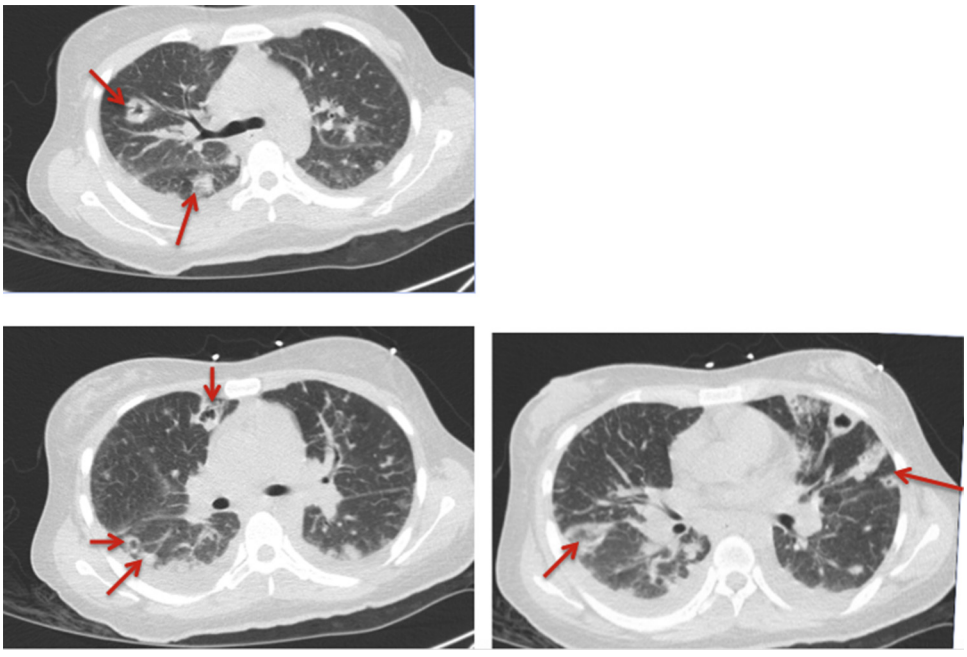

Fig. 1. Three chest commuted tomography (CT) scan images of the patient at presentation, showing multiple levels of the chest. There are bilateral, multifocal, cavitary nodules and consolidative opacities consistent with septic emboli involving upper and lower lung lobes bilaterally, and bilateral pleural effusions (full arrows).