

Massive Hemoptysis after Generalized Tonic Clonic Seizure Requiring Mechanical Ventilation

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A 38-year-old woman presented with massive hemoptysis (> 200 mL/24 hours) occurring abruptly after generalized tonic clonic seizure. She experienced similar episodes of hemoptysis on three later occasions. Although the coexistence of hemoptysis and seizure has been reported, albeit rarely, as a clinical manifestation of postictal neurogenic pulmonary edema, massive hemoptysis after seizure is an extremely rare event with no recurrent cases of such episodes having ever been reported. The coexistence of hemoptysis and seizure increases the difficulty in diagnosis for the clinician. We describe the differential diagnosis among the diseases capable of causing seizure and hemoptysis.

Key Words: Seizure, hemoptysis

INTRODUCTION

Massive hemoptysis is a common yet serious clinical condition which is typically caused by pulmonary tuberculosis, bronchiectasis and aspergilloma in Korea. Its occurrence after seizure is a rare clinical manifestation gave diagnostic difficulties to the clinician and is rarely reported in the clinical setting of neurogenic pulmonary edema (NPE).¹⁻⁵ We report a case presenting recurrent episodes of hemoptysis, consisting of two episodes of massive hemoptysis followed by two later episodes of non-massive hemoptysis after generalized tonic clonic seizure, and discuss the differential diagnosis of diffuse alveolar hemorrhage.

CASE REPORT

In March 17, 1998, a 38-year-old woman visited the emergency room presenting with massive hemoptysis (> 200 mL/24 hours) and dyspnea which occurred abruptly after generalized tonic clonic seizure. She denied smoking and drinking alcohol. She had no history of any other medical illnesses including heart diseases, tuberculosis or connective tissue disease. Because she was a shaman, her compliance to prescribed medication was poor. She had experienced a series of complex partial seizures 3 years previously. Those seizures had recurred about once a month with intermittent irregular antiepileptic medications. Repeated EEG recording showed intermittent left temporal spikes and brain MRI revealed a small lesion of a few millimeters in diameter/length, of unknown nature, in the left basal temporal cortex.

For 15 days prior to visiting the emergency room, she had ceased to take the valproic acid, which had been prescribed since her first partial seizure 3 years previously. Her typical seizure comprised a generalized heat flushing sensation followed by staring, lip smacking and loss of consciousness with secondary generalization. The clinical seizure diagnosis was complex partial seizure with secondary generalization, while the epilepsy diagnosis was left temporal lobe epilepsy. Her blood pressure was 140/80 mmHg, pulse rate 100 beats/min, respiratory rate 32/min and body temperature 37.2°C. In auscultation, coarse crackles were heard over the whole lung field and her heart beat was regular without murmur. Arterial blood gas study on room air revealed pH 7.38, PaCO₂ 38 mmHg, PaO₂ 38 mmHg, HCO₃⁻ 22 mEq/L and SaO₂ 76%. Labora-

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tory study confirmed whole blood cell count $6,700/\text{mm}^3$ (neutrophil, 79.7%), hemoglobin 11.5 g/dl, platelet count $256,000/\text{mm}^3$, aspartate aminotransferase 16 IU/L, alanine aminotransferase 12 IU/L, blood urea nitrogen 16.0 mg/dl, creatinine 0.8 mg/dl, prothrombin time 13.5 seconds (normal range, 11 - 15 sec.), activated partial prothrombin time 33 seconds (control, 29 - 44 sec.), creatinine kinase 150 IU/L (normal range, 30 - 165 IU/L) and MB fraction 1 IU/L. Urine analysis was normal. Sputum gram stain and culture for bacteria revealed a few polymorphonuclear neutrophil leukocytes and α -streptococcus as the predominant organism. Direct sputum examination produced negative results for acid-fast organisms and fungi. Chest X-ray showed diffuse infiltration on both lung fields. Her EKG showed normal findings. Echocardiogram revealed elevated right ventricular systolic pressure (RVSP): 41 mmHg with no regional wall motion abnormalities and normal left ventricular ejection fraction. The patient was intubated and mechanically ventilated with F_iO_2 100%. After 6 hours of mechanical ventilation, arterial blood gas study was as follows: pH 7.28, PaCO_2 44 mmHg, PaO_2 191 mmHg, HCO_3^- 20 mEq/L and SaO_2 99%. On fiberoptic bronchoscopy, scanty quantities of fresh blood were seen around both main and lobar bronchi, and no endobronchial lesion was observed. Hemosiderin-laden macrophages were not seen on the bronchial washing specimen. The results of bronchial washing and sputum examinations were negative for acid-fast organisms, fungi and bacteria. Studies for rheumatoid factor, ds anti-DNA, ANA and ANCA showed negative results. Valproic acid and empirical antibiotics were administered intravenously. No other medications, including corticosteroid or diuretics, were administered. The blood level of valproic acid was $15.6 \mu\text{g}/\text{mL}$ (therapeutic range: 50-100 $\mu\text{g}/\text{mL}$). On the 2nd day after admission, chest CT scan revealed diffuse, ground glass opacities on both lung fields (Fig. 1). On the 3rd day, the patient was weaned from the mechanical ventilator. The chest radiographic finding was significantly improved (Fig. 2). Repeated echocardiogram showed normal RVSP. She was discharged without any complaints of dyspnea or hemoptysis on the 7th day.

Similar clinical manifestations, including dys-

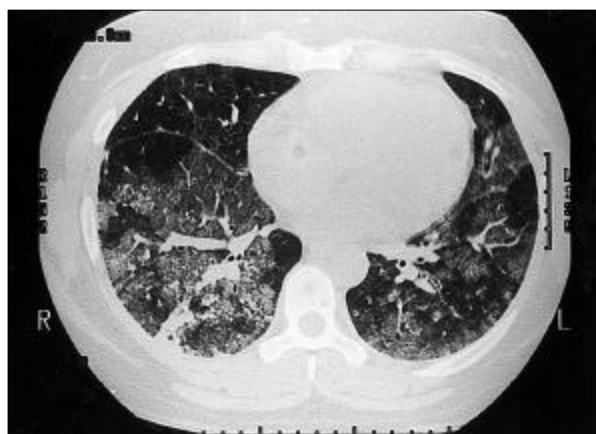


Fig. 1. Diffuse, ground glass opacities of both lung fields on chest CT scan at the 1st day after first admission.

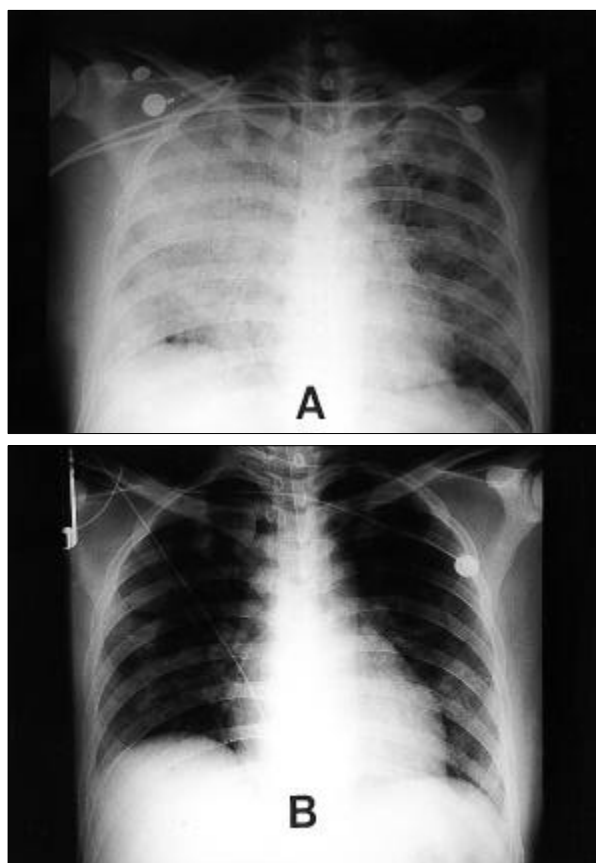


Fig. 2. Rapidly cleared, diffuse infiltrations on chest radiography between the 1st(A) and the 3rd(B) day after first admission.

pnea and hemoptysis within several minutes of each other followed by generalized tonic clonic seizure, were repeated on three more occasions, each time several days after the patient again

discontinued taking the anticonvulsive medication; massive hemoptysis in Aug. 1998, and non-massive hemoptysis in Mar. 1999 and Aug. 2001. Within one to two days of admission, the diffuse infiltrations on chest X-ray were cleared and the initially increased RVSP were returned to normal on serial echocardiography, merely with the recommencement of anticonvulsive medication, in combination with mechanical ventilation on one occasion and oxygen via mask on the other two occasions. In Aug. 2001, repeated brain MRI results were normal and no evidence of endobronchial lesions was seen on fiberoptic bronchoscopy. Studies for acid-fast organisms, fungi, bacteria and cytology were all negative.

DISCUSSION

Massive hemoptysis is a serious condition which requires prompt decision making from the clinician. Its etiology has been reported as pulmonary tuberculosis, bronchiectasis, lung abscess, mycetoma, lung cancer, and broncholithiasis.⁶ Because bilateral, diffuse, alveolar infiltration was observed on chest CT scan, diffuse alveolar hemorrhage syndrome and pulmonary edema were initially included in the differential diagnosis. In the presenting case, the absence of anemia and hemosiderin-laden macrophages on the bronchial washing specimen, the rapid improvement of clinical manifestation, and the relatively longer interval between episodes of hemoptysis excluded the clinical possibility of diffuse alveolar hemorrhage syndrome, and also cardiogenic pulmonary edema was excluded with the findings of transthoracic echocardiogram and the level of cardiac enzyme. We could further exclude the possible presence of isolated pulmonary capillaritis as a cause of diffuse alveolar hemorrhage because of the recurrent episodes of massive hemoptysis after the seizures, and the rapid improvement demonstrated on chest radiography without the use of immunosuppressive agents in the presenting case.⁷ Diffuse alveolar hemorrhage has been rarely reported in cases of malignancy or bronchial artery rupture, and both possibilities were easily excluded in the

presenting case.⁸⁻¹³

Considering the temporal relationship of the hemoptysis to the seizure, the postictal NPE may have been the possible hemoptysis cause. Since the first reported case of postictal NPE by Shanahan in 1908, only 43 cases have been reported in the world, none of which involved massive hemoptysis.^{1-5,14} The fact that all of these postictal NPE cases were reported with small amounts of hemoptysis after seizure and fine crackles on auscultation of the chest during the course of the disease may exclude the possibility of hemoptysis caused by postictal NPE in the presenting case which showed massive hemoptysis on two occasions and coarse crackles on both lung fields. Serial echocardiogram at the time of emergency room admission was performed on the presenting case. The dynamic clearing of pulmonary infiltration observed on serial chest radiography was associated with rapid normalization of the right ventricular systolic pressure on all four occasions of admission. The left ventricular function on echocardiogram and the level of cardiac enzyme were both normal. These findings suggest that pulmonary vascular hypertension after seizure, rather than increased pulmonary capillary permeability, may cause the sudden rupture of the pulmonary capillary and directly induce diffuse alveolar hemorrhage and massive hemoptysis.^{2,15-19} Pacht et al reported the first case of postictal NPE with gross hemoptysis.³ Although they did not clarify the level/severity/ degree of the sudden hemoptysis, gross hemoptysis may be caused solely by the sudden rupture of the pulmonary capillary, rather than by postictal NPE.

After excluding the possibility of postictal NPE, the following three cases with hemoptysis after seizure have been reported: one with necrotizing pneumonia caused by aspiration during seizure, another with lymphangioleiomyomatosis with tuberous sclerosis, and a third with cystic fibrosis.²⁰⁻²²

We report the first case of recurrent episodes of massive hemoptysis and diffuse alveolar hemorrhage after seizure, in a patient with no underlying lung disease.

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