



Cerebral Air Embolism and Cardiomyopathy Secondary to Large Bulla Rupture during a Pulmonary Function Test

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Cerebral air embolism combined with cardiomyopathy secondary to pulmonary barotrauma is rare. Here, we report an unusual case of cerebral air embolism combined with transient cardiomyopathy secondary to large bulla rupture during a pulmonary function test after lung cancer surgery. The patient experienced loss of consciousness. Computed tomography and magnetic resonance imaging suggested a cerebral air embolism. Electrocardiography showed ST-segment elevation and abnormally high plasma levels of cardiac enzymes. Echocardiography and coronary angiography suggested cardiomyopathy. The patient was discharged with no sequelae.

Keywords: Air embolism, Cardiomyopathies, Bullae, Barotrauma

Case report

A 63-year-old man with a history of hypertension and chronic kidney disease was admitted to the emergency department for acute loss of consciousness after performing the Valsalva maneuver during a pulmonary function test (PFT). His medications included candesartan, hydrochlorothiazide, and aspirin. Three months ago, he had received neoadjuvant chemoradiation therapy for lung cancer (adenocarcinoma, cT3N2M0, stage IIIB) in the right upper lobe; after a successful response, he received right upper lobe lobectomy (ypT1aN0M0) 11 days before the event.

On examination, his level of consciousness was semi-coma with no response to pain; right-side deviation of both eyeballs was noted, and both pupils measured 5 mm with a sluggish response to light. Initially, he had a blood pressure of 100/52 mm Hg, a heart rate of 102 beats/min, and a temperature of 36.8°C, and he received rapid-sequence intubation to protect his airway. Computed tomography (CT) scans of the brain revealed multifocal air density in the bilateral cerebral subarachnoid space and deep cerebral white matter. There was no evidence of brain hemorrhage. Magnetic resonance imaging of the brain revealed diffuse leptomeningeal enhancement and diffuse enhancement in the perivascular space of the bilateral deep white matter. Both imaging studies suggested cerebral air embolism (Fig. 1).

A chest radiograph showed a round area of consolidation in the right lower lobe lung field (Fig. 2B). Electrocardiography revealed sinus tachycardia at 130 beats/min, with ST-segment elevation of 2 to 3 mm in leads 2, 3, augmented vector foot, and V3 through V6 (Fig. 3A). Laboratory testing showed that his plasma levels of creatinine kinase (CK), creatinine kinase-muscle/brain (CK-MB), and troponin I were 83 U/L, 1.59 µg/L, and 0.01 µg/L, respectively. In a follow-up examination performed 3 hours after the event, electrocardiography showed sinus tachycardia at 117 beats/min and ST-segment elevation of 1 to 2 mm in leads 2 and V2 through V5 (Fig. 3B), and his plasma levels of CK, CK-MB, and troponin I were 156 U/L, 19.51 µg/L, and 4.70 µg/L, respectively. Transthoracic echocardiography (TTE) revealed a newly developed regional wall motion abnormality, which was not compatible with the coronary territories. The ejection fraction decreased from 62% to 41%. Emergent coronary angiography revealed neither coronary artery disease nor acute myocardial infarction lesions (Fig. 3C, D). In a follow-up examination 12 hours after the event, the patient's plasma levels of CK, CK-MB, and troponin I were 242 U/L, 31.21 µg/L, and 8.45 µg/L, respectively.

The patient was transferred to the intensive care unit. His white blood cell count and hemoglobin, platelet, glucose, ammonia, sodium, potassium, calcium, and magne-



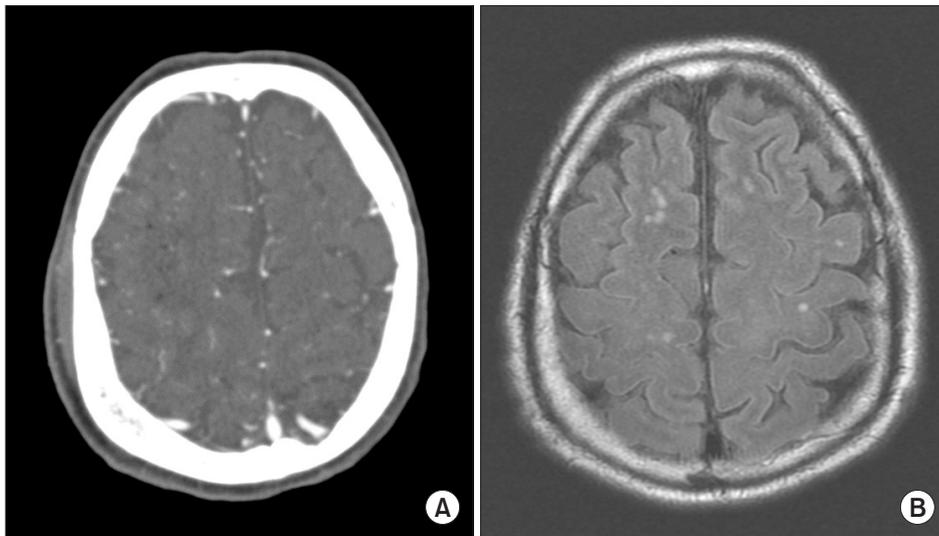


Fig. 1. (A) A computed tomography scan of the brain showed multifocal air density in the deep cerebral white matter, suggesting cerebral air embolism. (B) Magnetic resonance imaging of the brain showed diffuse enhancement in the perivascular space of the bilateral deep white matter in a T2 FLAIR image, suggesting acute cerebral infarction.

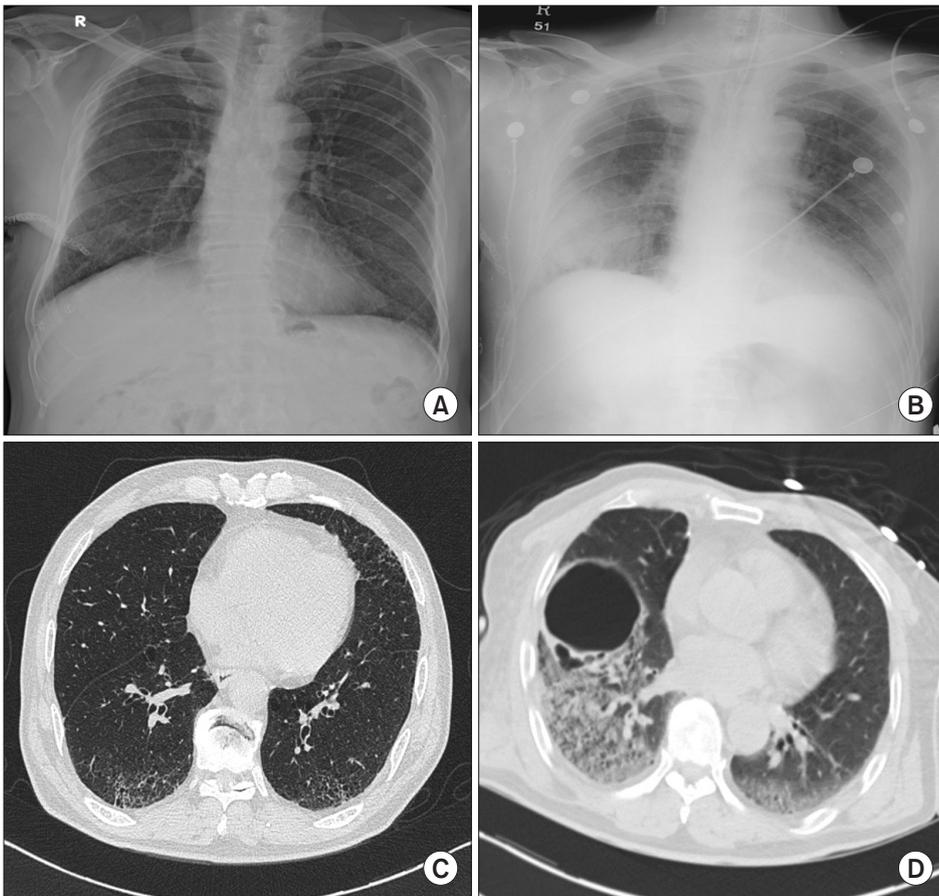


Fig. 2. (A) A chest radiograph before the pulmonary function test. (B) An initial chest radiograph after the patient lost consciousness shows a round consolidation, suggesting large bulla rupture. (C) A CT scan of the chest before right upper lobe lobectomy showed emphysematous lung, but no large bullae. (D) A CT scan of the chest 2 days after cerebral air embolism showed large bullae. CT, computed tomography.

sium levels were all within the normal range. In a follow-up examination 6 hours after the event, his level of consciousness was confusion, both pupils were 3 mm with a prompt response to light, and one step obey to command was pos-

sible. Generalized tonic-clonic seizures took place 11 to 20 hours after the event, and the seizures were controlled by an intravenous infusion of levetiracetam. On the first day after the event, electrocardiography showed sinus tachycar-

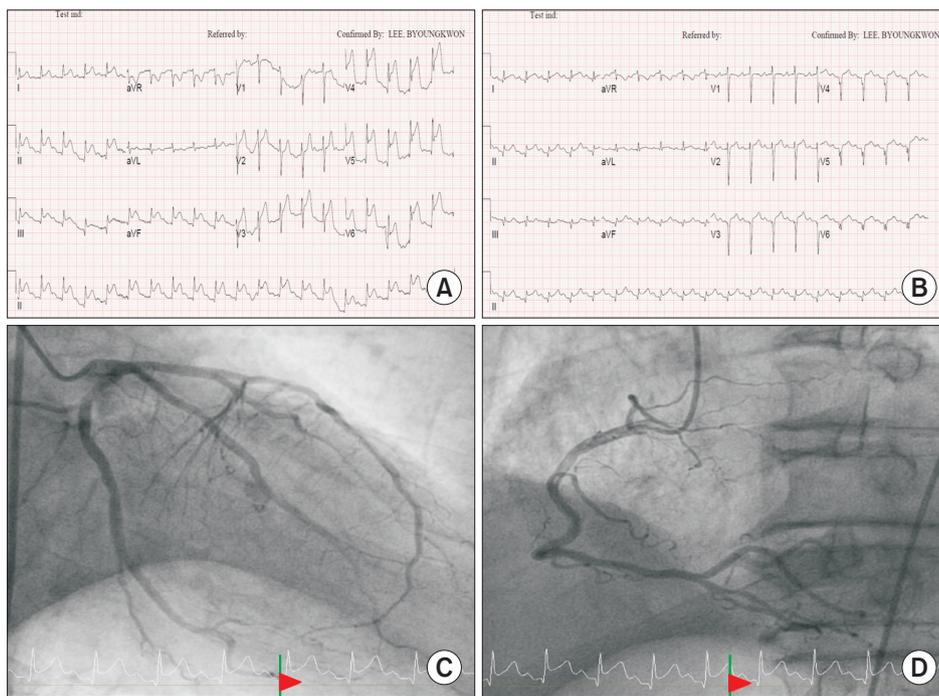


Fig. 3. (A) The initial electrocardiogram. (B) A 3-hour follow-up electrocardiogram. (C) Coronary angiography: left coronary artery. (D) Coronary angiography: right coronary artery. aVR, augmented vector right; aVL, augmented vector left; aVF, augmented vector foot.

dia at 107 beats/min with ST-segment elevation of 1 mm in leads V2 and V3, and his plasma levels of CK, CK-MB, and troponin I were 367 U/L, 13.51 $\mu\text{g/L}$, and 4.90 $\mu\text{g/L}$, respectively.

On the second day, the patient was alert, with chest radiography revealing a clear lung field. Mechanical ventilator weaning was performed. CT scans of the chest revealed a newly developed large bulla with a maximum diameter of 7 cm (Fig. 2D), and follow-up TTE showed an increased ejection fraction of 47%.

On the third day, the patient was transferred to the general ward. His plasma levels of CK, CK-MB, and troponin I were 1,642 U/L, 6.77 $\mu\text{g/L}$, and 1.12 $\mu\text{g/L}$, respectively. On the sixth day, electroencephalography revealed normal waveforms. On the seventh day, the patient was discharged without neurological symptoms or sequelae. Two weeks later, transesophageal echocardiography at the outpatient department revealed a grade I patent foramen ovale.

The patient provided written informed consent for the publication of clinical details and images.

Discussion

Cerebral air embolism secondary to pulmonary barotrauma is rare, although a few case reports have described cerebral air embolism secondary to bulla rupture during air travel or diving [1,2]. In these reports, the clinical pre-

sentations and outcomes ranged from benign to fatal. Our patient experienced cerebral air embolism secondary to bulla rupture, as in previous reports, but during PFT; furthermore, he presented with acute loss of consciousness, followed by seizures. Nonetheless, our patient was discharged with no severe sequelae.

Risk factors for bulla formation and rupture in our patient included emphysematous lung due a history of smoking, preoperative radiation therapy, fibrobullous changes in the ipsilateral residual lung after lobectomy, and a bleeding tendency due to aspirin. With regard to the first of those risk factors, Iwama et al. [3] showed a high frequency of emphysematous bullae in smokers, suggesting that smoking history is a risk factor for bulla formation. Our patient was an ex-smoker who quit 30 years ago and smoked 7.5 pack-years. Preoperative CT scans of the chest revealed diffuse pulmonary emphysema without large bullae. As a second risk factor, radiation-induced fibrosis may have occurred around the lung parenchyma during neoadjuvant chemoradiation therapy, making the bullae fragile to barotrauma, which led to rupture during the Valsalva maneuver. A total of 5,000 Gy of radiation was administered to the cancer mass in the right upper lobe and its margin. Intensity-modulated radiotherapy was used to minimize the dose to the surrounding normal tissue; however, fibrotic changes in the other lobe of the lung, including the right middle lobe and right lower lobe, were inevi-

table. With regard to the third risk factor, Tanaka et al. [4] reported fibrobullous changes in the lung parenchyma after lobectomy. They showed that the initial changes appeared at an average of 2 years (range, 3 months to 6 years) after lobectomy, with an incidence rate of 3%. They considered fibrobullous changes to be a late complication after lobectomy, but our patient developed fibrobullous changes only 3 weeks after lobectomy. This discrepancy may have been because our patient received radiation therapy preoperatively, whereas their study excluded patients who received preoperative and postoperative radiation therapy. Finally, the side effects of antiplatelet agents may result in bleeding around the bullae.

The direct cause of cerebral air embolism in this case was pulmonary barotrauma that occurred during PFT. The American Association for Respiratory Care (AARC) has suggested the following relative contraindications for spirometry: hemoptysis of unknown origin, unstable cardiovascular status, a recent myocardial infarction or pulmonary embolus, and recent thoracic or abdominal surgery [5]. Although the recommendations of the AARC are based on minimal concrete evidence [6], our patient had recently undergone lobectomy. Moreover, we suggest careful consideration of PFT in patients with pulmonary bullae. Farshchi Zarabi et al. [1] proposed 3 mechanisms of systemic air embolism secondary to bulla rupture: (1) direct embolization of the gas bubbles into the pulmonary veins and from there into the systemic circulation; (2) gas embolization into the pulmonary arterial system with subsequent incomplete filtration of the gas bubbles by the pulmonary capillaries; and (3) paradoxical embolization through a functional right-to-left shunt, such as a patent foramen ovale. Our patient was susceptible to all these possibilities.

Our patient also received a coronary evaluation due to ST-segment elevation and abnormally high levels of cardiac enzymes. Echocardiography and coronary angiography suggested stress-induced cardiomyopathy (SCMP). Any of the following mechanisms may lead to SCMP: direct air embolization into the coronary arteries, direct pressure on the coronary arteries and myocardium due to increased thoracic pressure during the Valsalva maneuver, and large swings in blood pressure during the Valsalva maneuver causing stress on the myocardium. To the best of our knowledge, our patient is the first case in which cardiac function and the coronary arteries were evaluated during cerebral air embolism secondary to bulla rupture. We suggest evaluating electrocardiography findings and plasma levels of cardiac enzymes in patients with cerebral air em-

bolism due to bulla rupture, especially in those at high risk of cardiovascular disease.

In conclusion, we presented a rare case of cerebral air embolism during PFT in a patient with a large pulmonary bulla. Cardiomyopathy was accompanied by cerebral air embolism secondary to pulmonary barotrauma for unknown reasons. In patients with bullous lung disease, mental status and electrocardiography monitoring are necessary during PFT, and medical practitioners should observe the patient carefully after PFT is completed. Follow-up chest radiography must also be done in bullous lung patients after PFT. Further studies are necessary to better address the relationship between cerebral air embolism secondary to pulmonary barotrauma and cardiomyopathy.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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