

Case of 78-Year-Old Male with Cerebral Gas Embolism Associated with Combined Pulmonary Fibrosis and Emphysema

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Cerebral gas embolism (CGE) from the thoracic cavity is commonly associated with invasive procedures, and cases of spontaneous CGE are rare. A 78-year-old man presented with severe spontaneous CGE associated with combined pulmonary fibrosis and emphysema (CPFE). To the best of our knowledge, the comorbidity of CGE in a CPFE patient has not been documented until now. The patient became unconscious with left hemiparesis at approximately 2 a.m. Computed tomography scan revealed minute air densities scattered in the deep white matter of the right frontal lobe. The patient died on the sixth day of hospitalization. We postulated that CGE can be classified as either arterial CGE or retrogradely infused venous CGE. To differentiate between these two, the distribution of air densities provided useful information in our case. We assumed that the air was infused through the pulmonary vein to the left atrium of the heart, resulting in arterial CGE. A review of the literature further suggests that an upright position at the time of a CGE attack might be related to the retrogradely infused venous origin of the air, whereas arterial CGE may more likely occur when the patient is lying down. **Key Words:** Cerebral gas embolism—artery—retrograde—venous—combined pulmonary fibrosis and emphysema.

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Case Report

Cerebral gas embolism (CGE) has been described as a decompression illness¹ and a complication of invasive procedures such as lung biopsy^{2,3} and central venous

catheterization.^{4,5} However, reports of spontaneous CGE associated with respiratory diseases are rare. Only cases of CGE associated with tuberculosis cavity,⁶ lung cancer,⁷ and post-transplant pneumonitis with positive pressure ventilation⁸ were reported. As far as we know, this is the first case report of spontaneous CGE associated with combined pulmonary fibrosis and emphysema (CPFE).

A 78-year-old man admitted unconscious to our hospital had left hemiparesis onset during sleep. He had a 4-year history of CPFE and frequent coughing. Brain computed tomography revealed scattered air-density areas in the deep and subcortical area in the right frontal lobe and a blurred border between the gray matter and the white matter, in the right middle cerebral artery region (Fig 1, A). Magnetic resonance imaging showed high-intensity areas on diffusion-weighted images, strongly

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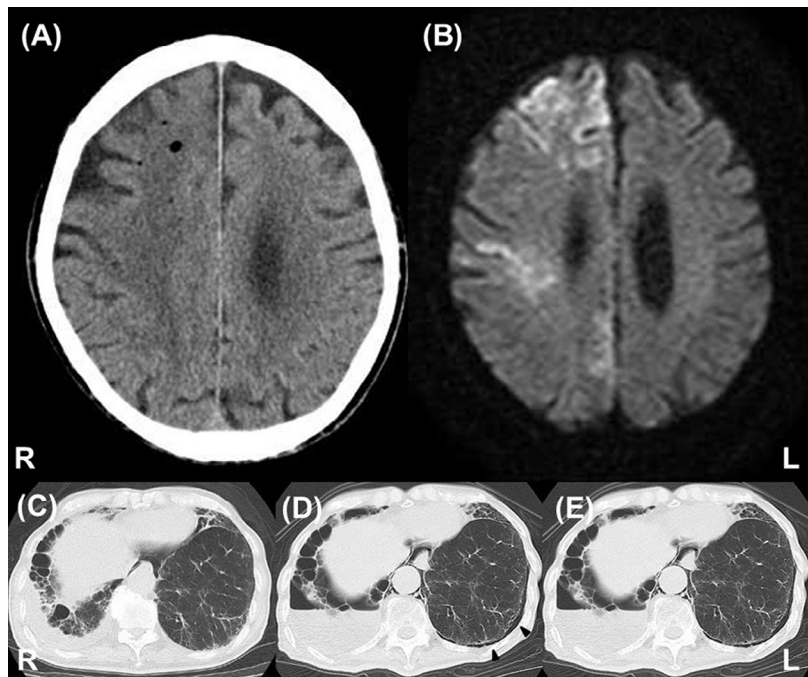
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Figure 1. (A, B) Brain images on admission. (A) CT scan. (B) Diffusion-weighted image (axial, 3.0 T; TR 500 ms; TE 92 ms; b value 1000 s/mm²). (C, D, E) Chest CT. (C) Three months prior to admission. (D) Two weeks prior to admission. (E) On admission. (A, B) Air densities found deep in white matter of right frontal lobe, and the blurred border between the gray matter and the white matter was seen. High-intensity areas on magnetic resonance image suggest acute cerebral infarction that is consistent with the findings on the CT image. (C, D, E) Three months prior to his admission, an emphysematous change and pleural effusion were found, as well as interstitial pneumonitis. Two weeks prior to admission, minor pneumothorax was noted around the left lung (arrowhead). On admission, the pneumomediastinum around the aorta, the deterioration of the pneumothorax in the left thoracic cavity, and fluid collection in the right were noted. The pleural effusion forms a niveau, suggesting some connection between the thoracic cavity and the external environment. Abbreviation: CT, computed tomography.



suggesting an acute cerebral infarction (Fig 1, B). Lung computed tomography demonstrated deterioration of the pneumothorax and pneumomediastinum (Fig 1, C-E). Contrast echocardiography using microbubbles showed no right-to-left intracardiac shunting. He died of respiratory dysfunction on day 6 of hospitalization. The patient's family did not give consent to an autopsy.

The presumed diagnosis was arterially infused CGE associated with CPFE. Scattered air-density areas in the frontal lobe suggests the migration of air into small perforated arteries,² and the areas of brain infarction indicated arterial, not venous, infarction. Probably, air first entered the pulmonary venous system through a bronchovenous or alveo-venous fistula in the pneumothorax or interstitial emphysema then translocated to the cerebral artery via the systemic circulation.³ The absence of intracardiac shunting further supports this opinion. In this case, coughing may have exacerbated the increase in the intratracheal pressure, triggering CGE. In contrast, air has been found in cerebral veins in other CGE cases with central venous catheterization.⁵ When air migrates into the jugular vein via the catheter, it may rise retrogradely to cerebral veins, particularly when patients are sitting upright. An upright position would facilitate retrograde flotation of air against gravity, as opposed to a lying position.

In conclusion, we report a patient with CGE associated with CPFE. Scattered air-density areas in the

hemisphere suggests a diagnosis of arterial CGE. His position at the time of the attack helped us differentiate arterial CGE from retrogradely infused venous CGE.

References

1. Tamaki H, Kohshi K, Sajima S, et al. Repetitive breath-hold diving causes serious brain injury. *Undersea Hyperb Med* 2010;37:7-11.
2. Azzola A, von Garnier C, Chhajed PN, et al. Fatal cerebral air embolism following uneventful flexible bronchoscopy. *Respiration* 2010;80:569-572. doi:10.1159/000321849.
3. Hiraki T, Fujiwara H, Sakurai J, et al. Nonfatal systemic air embolism complicating percutaneous CT-guided transthoracic needle biopsy: four cases from a single institution. *Chest* 2007;132:684-690.
4. Brouns R, De Surgeloose D, Neetens I, et al. Fatal venous cerebral air embolism secondary to a disconnected central venous catheter. *Cerebrovasc Dis* 2006;21:212-214.
5. Lai D, Jovin TG, Jadhav AP. Cortical vein air emboli with gyriform infarcts. *JAMA Neurol* 2013;70:939-940. doi:10.1001/jamaneurol.2013.1949.
6. Ferry T, Argaud L, Delafosse B, et al. Inactive tuberculosis cavity responsible for fatal cerebral air embolism. *Intensive Care Med* 2006;32:622-623.
7. Otomune K, Yamamoto S, Kohara N. Cerebral air embolism associated with lung cancer. *Intern Med* 2011; 50:2439.
8. Hung SC, Hsu HC, Chang SC. Cerebral air embolism complicating bilevel positive airway pressure therapy. *Eur Respir J* 1998;12:235-237.