Cerebral air embolism as a complication of idiopathic pulmonary fibrosis. A case report.

N. Brunelli[3] MD, C. Roberti[1] MD, D. Pantoli[2] MD, A. Carnevale[1] MD, V. Giuliani[2] MD, E. Ferraro[1] MD, L. Polidori[1] MD, M. Santarelli[1] MD, M. Paolucci[3] MD, A. Cascio Rizzo[3] MD, G. Di Battista[1] MD.

[1]Neurology Unit and [2]Neuroradiology Unit, San Filippo Neri Hospital, Rome
[3]Neurology Unit, Campus Bio-Medico University, Rome.

INTRODUCTION

Cerebral air embolism (CAE) is a rare cause of stroke and may be caused by iatrogenic[1,2] or traumatic introduction of air into the arterial or venous circulation. There are few reports on CAE occurring in non-iatrogenic situations.

CASE PRESENTATION

A 79-year-old man with a medical history of pulmonary fibrosis with pulmonary hypertension was admitted to our hospital for acute onset of facial droop followed by loss of consciousness. On neurologic examination, he was stuporous, had miotic pupils and a coniugate eye deviation to the right, verbal output was unintelligible, had limb hypotonia with some purposeful movements to noxious stimuli only on right limbs. CT angiogram of the brain revealed air bubbles along the sulci in parietal area, mainly on the right. Chest CT scan showed, in addition to the known honeycomb lung, a small pneumothorax of the right apex and a pneumomediastinum. EEG suggested a structural lesion in the right hemisphere. On day 2, head CT scan revealed no air emboli. After few days, the patient recovered consciousness and showed a left brachial-crural plegia. Brain MRI, performed a week after the onset of symptoms, revealed bilateral high DWI signal in cortical-subcortical parietal-occipital area, with contrast enhancement predominantly on the right (Figure 1). Pneumothorax and pneumomediastinum spontaneously healed after some days and the patient was sent to a neuro-rehabilitation structure.

DISCUSSION

It has been reported that CAE shows several characteristics on brain imaging, which may be useful for diagnosis. The most common site of CAE is the watershed area, especially the border zone of the middle and anterior cerebral arteries[3]. Moreover, usually, CAE shows restricted infarct patterns on DWI sequences, showing strong hyperintensity throughout the cortical gray matter, in a “multiple gyriform” pattern.[3,4] In our case, air emboli have been located in the border zone of middle and posterior cerebral arteries.

Another characteristic of the present case is the small pneumothorax with pneumomediastinum. CAE with pneumomediastinum is rare; we found only two reports in the literature. Matsuura et al[5] reported a case of a CAE complicated by pneumomediastinum, which occurred during eating, in a patient with suspected interstitial pneumonia. Tabata et al[6] reported a case of CAE caused by pneumomediastinum after vomiting in a patient with chronic interstitial pneumonia. Our chest CT also revealed pulmonary fibrosis with honeycomb lung. The association between pneumomediastinum and interstitial pneumopathy has been mentioned in different articles.

Franquet et al[7] found spontaneous pneumomediastinum in 4 of 78 patients (5.1%) with idiopathic pulmonary fibrosis. The mediastinal pathophysiology is attributed to the Macklin effect: an alveolar rupture due to an increase in intrathoracic pressure, followed by air dissection through the bronchovascular sheath into the mediastinum.[8] The definite mechanism how the air enters into the vessels in absence of any invasive manipulation is unclear. Edwardson et al[9] reported several cases of CAE during air travel. In those reports, the patients had the complications of bronchogenic cysts. Probably, expansion of the cyst is induced by the air pressure change in the airplane after takeoff, which results in subsequent disruption of alveoli and the surrounding tissues including blood vessels. A similar mechanism was hypothesized in a case of CAE caused by the inhalation of helium.[10] The uncontrolled delivery of helium directly from the high-pressure tank into the patient’s airway generated sufficient transpulmonary pressure to rupture alveoli and the surrounding blood vessels, thus introducing helium into the pulmonary veins, then into left heart and finally into the systemic arteries as into the cerebral arteries causing stroke.

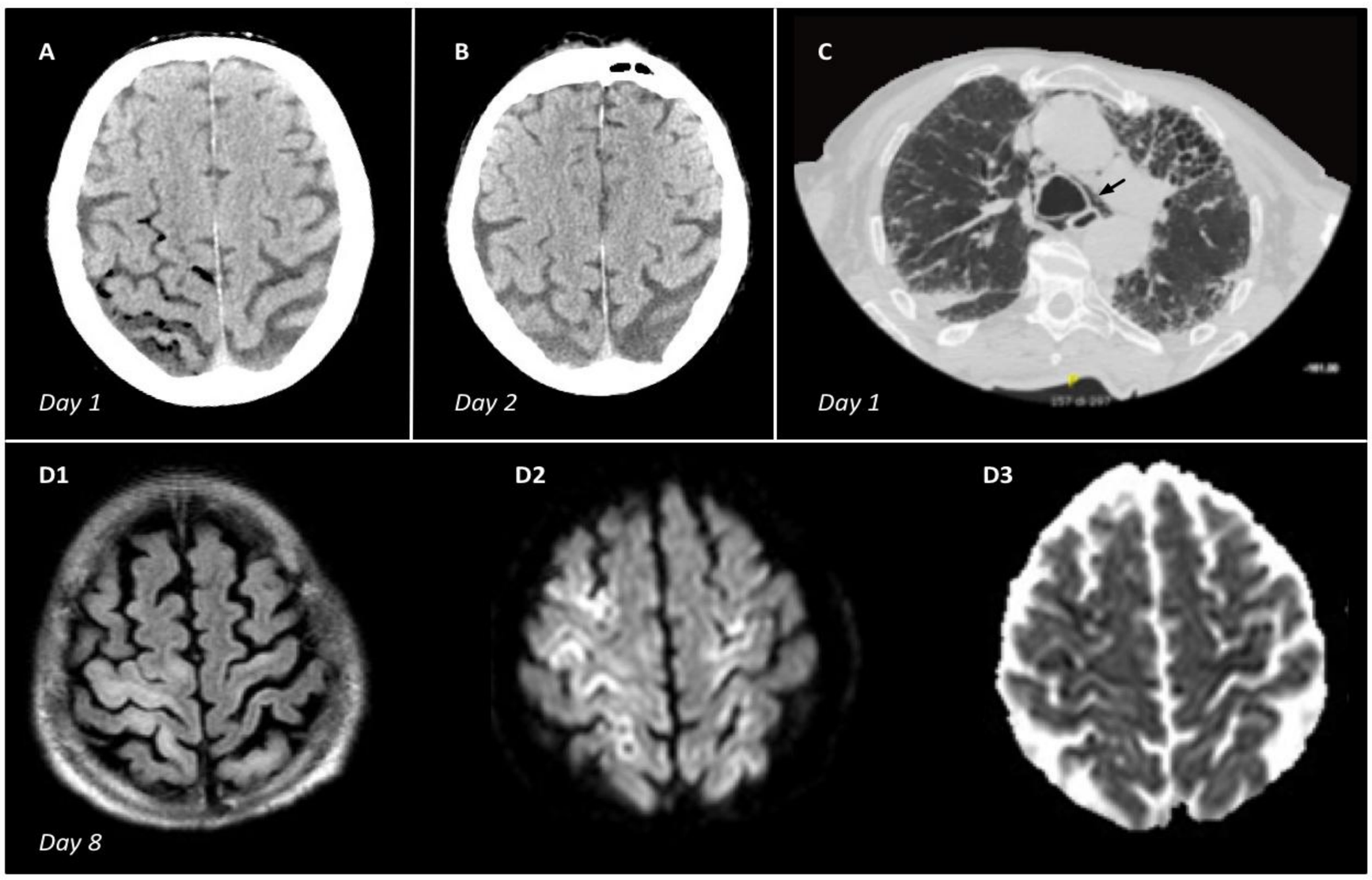

Figure 1 - (A) Head CT scan performed on day 1. (B) Head CT scan performed on day 2. (C) Chest CT scan performed on day 1 showing pulmonary fibrosis, bronchiectasis and pneumomediastinum (arrow). (D1-D2-D3) Brain MRI performed on day 8: (D1) FLAIR imaging of magnetic resonance; (D2) DWI imaging; (D3) ADC imaging.

CONCLUSION

Our report confirms that CAE can occur in a non-iatrogenic situation, especially in a patient with pulmonary vulnerability. To our knowdlege, this is the first case of CAE that occurred spontaneusly in a patient with an underlying idiopathic pulmonary fibrosis. Pulmonary hypertension might have caused functional opening of an anatomic shunt with transpulmonary air passage.

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CAMPUS BIO-MEDICO UNIVERSITY OF ROME
Via Álvaro del Portillo, 21 - 00128 Rome - Italy
www.unicampus.it