ANEURYSMAL SUBARACHNOID HEMORRHAGE PRESENTING WITH NEUROGENIC PULMONARY EDEMA AND HEMOCONCENTRATION

Hung-Lin Hsu MD*; Chien-Chin Hsu, MD, PhD**,***; Kuo-Tai Chen, MD*,***

* Emergency Department, Chi-Mei Medical Center, Tainan, Taiwan

** Department of Biotechnology, Southern Tainan University of Technology, Tainan, Taiwan

*** Department of Emergency Medicine, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

Abstract

Introduction: Neurogenic pulmonary edema (NPE) is a life-threatening condition caused by various significant central nervous system injuries. It is characterized by an abrupt increase in fluids within the alveoli and interstitium of the lungs. The mechanism underlying NPE remains unclear. Few pathophysiological causes-including neurocardiac factors, neurohemodynamic factors, blast theory, and pulmonary venule adrenergic hypersensitivity-have been proposed

Case report: We report the case of a patient with NPE associated with subarachnoid hemorrhage (SAH) and stress-hemoconcentration. Adrenergic responses have been reported to play a crucial role in both NPE and stress-hemoconcentration. We believe that SAH induces an adrenergic surge, which causes a cascade of reactions and leads to both NPE and stress-hemoconcentration.

Conclusion: Central nervous system (CNS) injuries are crucial and common causes of adrenergic stimulation; therefore, any subtle signs indicating CNS injuries should be carefully investigated. The distress of NPE may mask the initial symptoms of CNS injuries. If the patient is intubated and under mechanical ventilation to treat NPE, the inciting neurological injury may be overlooked and result in a delayed diagnosis.

Keywords: subarachnoid hemorrhage; neurogenic pulmonary edema; hemoconcentration; stress; adrenaline

Introduction

Neurogenic pulmonary edema (NPE) is characterized by the development of acute pulmonary edema shortly after a central nervous system (CNS) injury, including subarachnoid hemorrhage (SAH), intracranial hemorrhage, traumatic brain injury, status epilepticus, and spinal cord injury(1,2). In most case reports, the symptoms of neurogenic insults have been stated to precede the development of acute pulmonary edema. This unique clinical manifestation indicates to healthcare workers that pulmonary edema results from strong CNS injury, thus making the diagnosis of NPE reasonable in patients with acute pulmonary edema.

We report the case of a 52-year-old man who presented to the emergency department with acute pulmonary edema and hemoconcentration without any inciting etiology. Symptoms of cerebral aneurysmal SAH developed 4 hours after presentation. This unusual presentation reminded the emergency physicians that, although rare, excess adrenergic stimulation caused by a CNS injury is a possible cause of unexplained acute pulmonary edema. Severe respiratory distress due to acute pulmonary edema may mask the initial symptoms of CNS injuries.

The accompanying hemoconcentration observed in our patient subsided after the aneurysmal SAH was treated. Herein, we attempt to explain the pathogenic mechanisms of NPE and hemoconcentration due to stimulation by a CNS injury such as SAH.

Case report

Our patient was a 52-year-old man with a history of hypertension who received antihypertensive therapy regularly. He presented at our hospital because of a sudden onset of shortness of breath in his sleep. On arrival at the emergency department, his vital signs were as follows: temperature 36.3°C; pulse rate 92 beats/min; respiratory rate 26 breaths/min; arterial pressure 213/135 mm Hg; and oxygen saturation of 89% (determined by pulse oximetry). He was in a state of

respiratory distress with clear consciousness. A physical examination revealed cyanotic lips, rales on both lung fields, and cold and diaphoretic skin without limb edema or engorgement of the jugular vein. The neurological examinations of the patient are normal on admission. He did not report chest pain, fever, or cough. After the patient received 100% oxygen supplement using a nonrebreathing mask, his respiratory distress was alleviated and his oxygen saturation was increased to 95%. A roentgenogram of the chest revealed a moderate-sized heart and bilateral pulmonary edema (Fig. 1). Laboratory tests revealed normal levels of C-reactive protein (0.4 mg/L). Renal function tests (creatinine: 1.02 mg/L) showed normal results, and the levels of cardiac enzymes (troponin I: 0.0056 µg/L) and B-type natriuretic peptide (0.146 µg/L) were normal. In addition, a complete blood cell count revealed hemoconcentration, with a white-cell count of 16300 cell/µL, hemoglobin level of 25.7 g/L, and platelet count of 614000 cell/µL. The emergency physician prescribed intravenous nitroglycerin and furosemide and performed a phlebotomy to release 500 mL of blood. The patient's symptoms further improved.

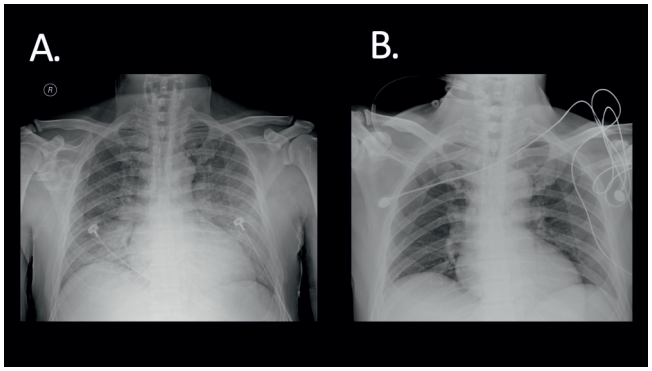

FIGURE 1. A. Roentgenogram of the chest on admission revealed bilateral pulmonary edema. B. Forty days after admission, the pulmonary edema had resolved.

After 4 hours, he felt pain in the posterior region of his neck and experienced left eye ptosis. A cerebral computed tomographic scan revealed SAH in the basal cisterns and fissures (Fig. 2). Subsequent cerebral computed tomographic angiography revealed a ruptured aneurysm in the left internal carotid artery (Fig. 3). An echocardiogram of the patient revealed left ventricular hypertrophy, adequate contractility (left ventricular ejection fraction: 73.8%), and no regional wall abnormality. The patient underwent endovascular stenting for the ruptured aneurysm on the fourth day after admission. His pulmonary edema and hemoconcentration resolved spontaneously the next day (white-cell count of 5500 cell/µL, hemoglobin level of 14.7 g/L, and platelet count of 338000 cell/µL).

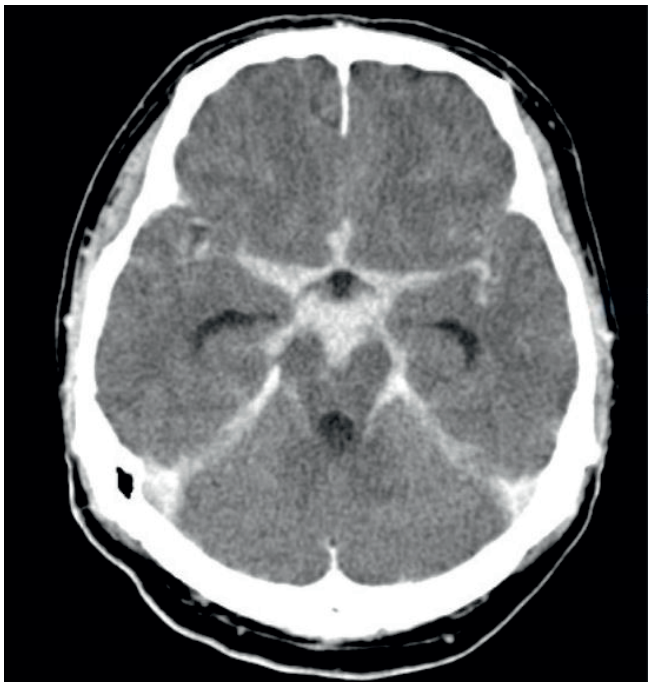

FIGURE 2. Cerebral computed tomographic scan revealed subarachnoid hemorrhage

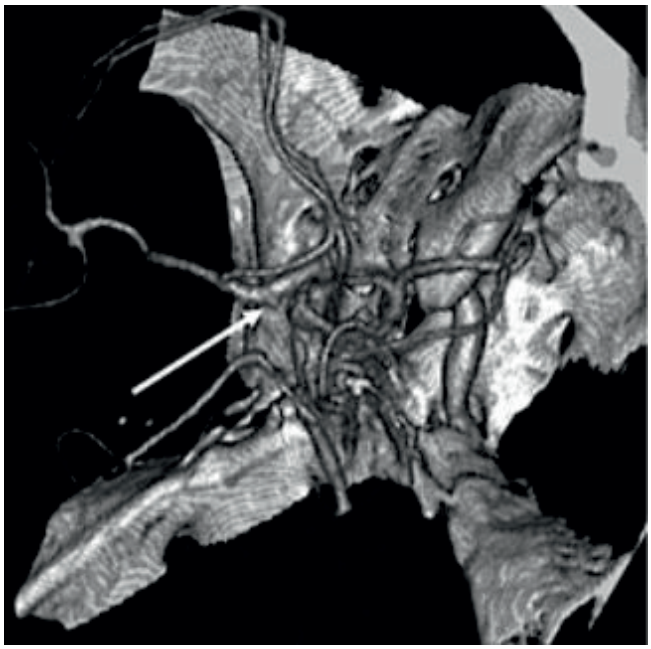

FIGURE 3. Cerebral computed tomographic angiography revealed a ruptured aneurysm on the left internal carotid artery (arrow: left internal carotid artery aneurysm)

After 50 days in hospital, he was discharged with left hemiparesis and clear consciousness. He did not require specific therapies for pulmonary edema or hemoconcentration during outpatient follow-up. Written informed consent couldn't be obtained due to the impossibility of reaching the patient.

Discussion

Davison et al. proposed four pathogenic mechanisms-namely neurocardiac factors, neurohemodynamic factors, blast theory, and pulmonary venule adrenergic hypersensitivity-responsible for the development of NPE(2). Additionally, Baumann et al. reported that the effects of inflammatory mechanisms due to a major neurologic injury may contribute to the development of NPE(1). In the case of our patient's NPE, we believe that the "blast theory" mechanism and the "inflammatory mechanism" both played crucial roles. A sudden increase in the intracranial pressure induced an α-adrenergic response, which caused obvious pulmonary vasoconstriction as well as altered vascular permeability. The "blast theory" mechanism involved a high-pressure hydrostatic influence and pulmonary endothelial injury(2,3). Additionally, the "inflammatory mechanism" indicates that a major neurologic injury caused an inflammatory reaction and produced various cytokines, which induced an increase in the permeability of the pulmonary capillary, thus causing pulmonary edema(1).

The two aforementioned mechanisms are further supported because of the patient's concurrent hemoconcentration. Acute stress, such as a CNS injury, usually stimulates the adrenergic system and increases the blood pressure, which causes a net efflux of plasma into the interstitial spaces. This condition has been referred to as "stress-hemoconcentration"(3).

Furthermore, brain injury activates the sympathetic nervous system and releases cytokines, which damage the vascular endothelium and increase capillary permeability(4). These mechanisms are similar to the theories regarding the occurrence of NPE. Choi et al. reported a similar case in which a ruptured cerebral aneurysm was accompanied by an increase in all blood cell counts; the hematologic abnormalities normalized after treatment of the ruptured aneurysm(5).

In our case, the patient presented to the emergency department with unexplained acute pulmonary edema. His B-type natriuretic peptide level, renal function test results, and echocardiography results were all within the normal range, thus excluding the possibilities of intravascular fluid overload, renal failure, and impairment of cardiac contractility. Additionally, tachycardia and elevation of arterial pressures were recovered simultaneously. Excessive stimulation of the adrenergic system could be the etiology of acute pulmonary edema. The new onset hemoconcentration further raised the suspicion of excessive adrenergic activation.

Conclusion

CNS injuries are crucial and common causes of adrenergic stimulation; therefore, any subtle signs indicating CNS injuries should be carefully investigated. The distress of NPE may mask the initial symptoms of CNS injuries. If the patient is intubated and under mechanical ventilation to treat NPE, the inciting neurological injury may be overlooked and result in a delayed diagnosis.

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