Delayed Neurogenic Pulmonary Edema Resulting from a Ruptured Large Distal Anterior Cerebral Artery Aneurysm: A Case Report

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Summary: A 64-year-old patient presented with subarachnoid hemorrhage (SAH) due to ruptured right distal anterior cerebral artery aneurysm (DACA), which was classified as large size. In addition to the rarity of this aneurysm, this patient had delayed neurogenic pulmonary edema (NPE), which is atypical in view of the onset of this systemic complication of SAH. In general, NPE develops immediately after the ictus. Since this patient also suffered from other serious systemic complications such as disseminated intravascular coagulation (DIC) and liver dysfunction, we chose interventional procedures followed by intensive treatments to improve his poor status. In spite of these treatments he became severely disabled. In this paper, we discuss not only the rarity of a case with large ruptured DACA and delayed NPE but also the significance of appropriate management of the poor-grade patient with SAH.

Key words:
- delayed neurogenic pulmonary edema
- distal anterior cerebral artery aneurysm
- poor-grade subarachnoid hemorrhage
- vasospasm

Introduction

NPE is generally considered to be related to damage to the hypothalamus due to serious intracranial diseases. Although NPE developing immediately after an ictus or trauma and its mechanism have been discussed in many reports, delayed NPE has been described only in a few reports. We present a case of poor-grade subarachnoid hemorrhage (SAH) due to large ruptured right DACA. Apart from this uncommon event, this patient suffered delayed NPE which appeared about 31 hours after the attack. In this paper, we discuss the management of the patient with poor-grade and unusual clinical features (mainly delayed NPE) and review other related reports.

Case Report

A 64-year-old male was admitted to our hospital in a state of unconsciousness. Hypertension was noted but not treated. About 20 years previously this patient suffered from tuberculosis and was treated successfully. He had no previous history of cardiac failure. On admission his consciousness level was at Glasgow Coma Scale (GCS) 9 (E2 V3 M4). He manifested left hemiparesis (paralysis). Computed tomography (CT) revealed SAH (Fisher group IV). The clinical grade of SAH based on Hunt & Kosnik (H & K) classification was to IV. Thick hematoma was noted particularly in the interhemispheric fissure (Fig. 1). Angiography performed on admission revealed a large aneurysm (12×14×13 mm) at the supracallosal portion of right pericallosal artery (Fig. 2). This aneurysm was observed in the late venous phase (Fig. 4 left, center). Chest radiography performed about 2 hours after admission showed no significant findings to suggest pulmonary edema (Fig. 2 left). About 31 hours after the attack, this patient became increasingly tachypneic and cyanotic. However, the neurological signs were unchanged. Blood gas analysis with administration of oxygen (3 l/min) showed a PaO₂ of 68.3 mmHg, PaCO₂ of 35.0 mmHg, and

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pH of 7.473. Chest radiography revealed cardiomegaly and alveolar infiltrations located at the hilum suggestive of NPE (Fig. 2 right). For two days after admission, the fluid exceeding 3,000 ml/day was administered to the patient as hypertensive hypervolemic hemodilution therapy to prevent vasoconstrictions. Diuretics were administered carefully in order to avoid fluid overload. The urinary volumes on admission and the next day were 1,300 and 2,800 ml/day, respectively. Three days after admission, embolization of the right pericallosal artery, but not of the aneurysm, using interlocking detachable coils (IDCs) was performed (Fig. 4 right). On the same day, tracheostomy was performed and his respiration was assisted with a mechanical ventilator for 13 days. His cardiovascular function was monitored using a Swan-Ganz catheter. Pulmonary capillary wedge pressure (PCWP) often exceeded 20 mmHg of monitoring. Alveolar-arterial oxygen tension difference (AaDO₂) was not monitored. His blood pressure was controlled at about 150–160 mmHg (systolic pressure) by infusing nisopropine. During the course of treatment, his cardiovascular function ranged from I to II according to the classification of Forrester. About 14 days after admission, blood gas analysis and chest radiographic findings were almost normalized. He was then weaned from the mechanical ventilator. Postoperative angiography revealed obliterations not only of the aneurysm but also its parent artery (Fig. 4 right). CT revealed low-density areas irrigated by distal portions of bilateral pericallosal arteries (Fig. 5). Intracranial pressure (ICP) was monitored continuously until the Cerebrospinal fluid (CSF) drainage tube was extubated. The pressure mainly stayed below 20 mmHg but sometimes exceeded this level. CSF drainage tube was
inserted by ventriculostomy and remained for 24 days from the 3rd day after admission. Although a ventriculoperitoneal shunt to replace the CSF drainage tube was put in place about one month after admission, the patient was severely disabled with left hemiparesis and paraplegia.

**Discussion**

We presented this case because of extreme rarity in terms of the two points described below.

Initially, we would like to emphasize the rarity of the DACAA located at the supracallosal portion in our patient. Most anterior cerebral artery aneurysms arise at the bifurcation of the callosomarginal artery. In addition to the location of the aneurysm, a large aneurysm is extremely rare. To our knowledge, only four cases have been reported including ours.

We chose embolization of the parent artery but not of the aneurysm using (IDCs) as the management procedures. Since this aneurysm was observed in the late venous phase, and based on its shape, it may have been a pseudoaneurysm such as dissecting one. If this was indeed a pseudoaneurysm, coil embolization of the aneurysm is very dangerous and occlusion of the parent artery is therefore preferable. On the other hand, we could not carry out open direct surgery at the acute stage because of the critical neurological and systemic conditions of the patient.
order to prevent rebleeding of the aneurysm and administer intensive care for his critical condition, we could not choose an alternative management. Moreover, we must take the subsequent vasospasm into consideration. The subsequent optimum following therapy is best performed after aneurysm obliteration. Even in patients with poor grade SAH, good outcomes could be achieved with aggressive surgical and medical intervention in the early stage. We would like to receive criticism and suggestions from the readers about the treatments we carried out for this patient if possible.

Secondly, we stress that delayed NPE is also extremely rare. In general, NPE following SAH develops immediately after the ictus. Only a few reports have discussed this uncommon phenomenon. The definite diagnosis of NPE was not necessarily based only on radiographical findings. We must consider not only the alveolar infiltrations but also other parameters such as PCWP, Forrester's classifications, and the levels of C-reactive protein (CRP). Signs of inflammation, including elevation of the CRP levels, were noted in the present case and we could ignore the possibility of pneumonia. Moreover, fluid overload was suggested in this patient discussed as below. However, sufficient diuresis was observed for two days after admission. Based on these findings, we diagnosed the patient as having delayed NPE.

It is well known that pulmonary function is significantly related to the grade of the patient based on H&K classification on admission. The clinical grades according to H&K classification of the patients reported by Fisher et al. and Miyamori et al. were IV and V, respectively. The clinical grade of our case was IV. It is also widely accepted that NPE is closely associated with damage to the hypothalamus which may be induced by the rise in ICP due to SAH. This rise in ICP triggers widespread autonomic responses, particularly sympathetic vasomotor overactivity, resulting in immediate changes in the systemic and pulmonary circulations.

However, the mechanism underlying delayed NPE has scarcely been discussed previously. Miyamori et al. stated that delayed NPE may be related to the permeability of pulmonary vessels, but they did not describe the mechanism in detail. Fisher et al. commented that the cause of the delay of onset was not clear.

Hypertensive hypervolemic hemodilution therapy performed upon admission may be related to delayed NPE. More than 3,000 ml was administered to the case of Miyamori et al. for two days after admission. Our case similarly received more than 3,000 ml per day upon admission. However, sufficient diuresis was observed in the present case. There was also no evidence of fluid overload in the patient of Fisher et al. Therefore, this therapy may have induced the delayed NPE to some extent but it cannot be confirmed whether the therapy can cause the unusual state. We would like to obtain any available information about delayed NPE from the readers.

In conclusion, the exact mechanism underlying delayed NPE still remains unknown. However, we must keep in mind this unusual state which can develop several days after the ictus whenever we treat a patient with poor-grade SAH.

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**Abbreviations**

AaDO₂, alveolar-minus-arterial oxygen tension difference; CRP, C-reactive protein; CSF, cerebrospinal fluid; CT, computed tomography; DACAA, distal anterior cerebral artery aneurysm; DIC, disseminated intravascular coagulation; GCS, Glasgow Coma Scale; GOS, Glasgow Outcome Scale; GR, good recovery; H & K, Hunt & Kosnik; ICP, intracranial pressure; IDC, interlocking detachable coil; Jpn, Japanese; MD, moderately disabled; min, minute; NPE, neurogenic pulmonary edema; PCWP, pulmonary capillary wedge pressure; SAH, subarachnoid hemorrhage; SR, severely disabled; VS, vegetative state

**References**


**要 旨**

Large distal anterior cerebral artery aneurysm破裂に伴うくも膜下出血が誘因となった遅発性神経原性肺水腫の1例

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前大脳動脈瘤を発症した、一般に之部に発生し、末梢部いわゆるsupracallosal portionに発生するものはまれである。そのなかで大脳動脈瘤（最大径12mm以上）はさらに頻度が少なく、われわれの症例を含め、経験する例は4例のみであった。

一方、くも膜下出血に伴う神経原性肺水腫は、発症直後に出現する例が一般的であり、発症後12時間以上経ってからのものは遅発性神経原性肺水腫と呼ばれ、ほとんど報告がなく文献的考察もなされていない。

今回われわれは、上述の特異な2項目を有するくも膜下出血の症例を経験した。原因となった動脈瘤は、画像所見から解離性脳動脈瘤が示唆され、全身状態がきわめて不良であったため、血管内治療を選択した。これに加え、全身管理においては種々の集学的治療を行ったにもかかわらず、患者は高度の脱離症状を残した。

われわれは遅発性神経原性肺水腫の原因として、紙者の過度負荷を疑い、最終的に原因は同定できず、有用な文献的報告も得られなかった。今後著者も含め、脳神経外科医が遭遇しうる重要な病態であると考えられたため、問題提起の意味を使って報告した。

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