# Two Medullary Hemorrhage Cases Complicated by Respiratory Distress in the Early Phase

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Medullary hemorrhage is quite rare among brain stem hemorrhage cases, thus the clinical course remains unclear. In the medulla oblongata, respiratory centers are located and previous reports indicate that medullary lesions have possible relationship with acute respiratory distress syndrome. This kind of respiratory failure is commonly caused by neurogenic pulmonary edema (NPE), which is defined as noncardiac noninfectious acute respiratory distress syndrome with changes in intracranial condition including cerebrovascular events. However, to date, very few reports have described cases with medullary hemorrhage accompanied by NPE. We experienced 2 patients with medullary hemorrhages. A 65-year-old man presented with sudden onset of headache, whose head computed tomography showed right medullary hemorrhage. Another 76-year-old woman was transferred because of sudden limb weakness and diagnosed with left medullary hemorrhage. Digital subtraction angiography showed the presence of arteriovenous fistula in the medulla oblongata and drainer veins in the second case. Both cases were complicated by acute pulmonary edema in the early phase, suggesting the possible association of the medullary hemorrhage with NPE.

**Key Words:** Medullary hemorrhage—arteriovenous fistula—acute respiratory distress syndrome—neurogenic pulmonary edema



## Introduction

Medullary hemorrhage is 1 of the most uncommon types of strokes and the clinical details in this status remain unknown. Here we describe 2 medullary hemorrhage cases complicated by acute respiratory distress syndrome in the acute period.

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## Case Report

In the first case, a 65-year-old Japanese male with chronic atrial fibrillation had been administered warfarin. He was admitted after experiencing sudden onset of headache. The neurological examination suggested Wallenberg syndrome, and brain computed tomography indicated a right-side medullary hemorrhage (Fig 1A). On the first hospital day, he developed acute respiratory distress syndrome (ARDS) and intubation was performed (Supplemental Figure). Neurogenic pulmonary edema (NPE) was suggested, and thereafter he showed a good clinical course. The second case was a 76-year-old Japanese female with no significant medical history, who complained of sudden weakness in the right extremities. Head computed tomography showed a high-density area in the left dorsal medulla (Fig 1B). She also presented with severe desaturation that required temporal intubation. Brain magnetic resonance imaging (Fig 1C) and digital subtraction angiography (Fig 1D, E) indicated an

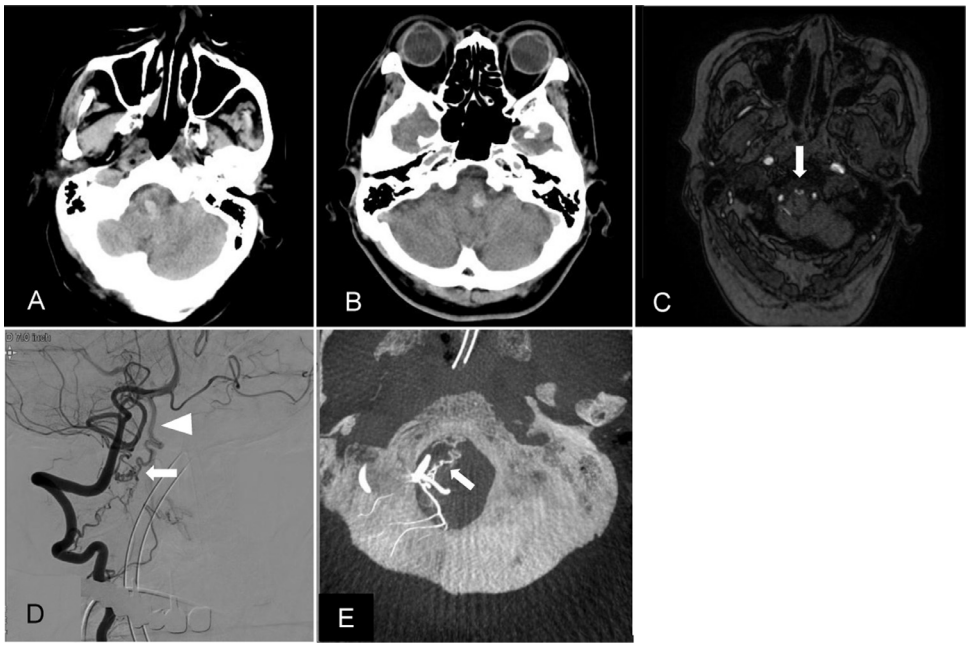


**Figure 1.** *Imaging findings. Case 1: Head CT (A) shows a left dorso-lateral hemorrhage. Case 2: Head CT (B) demonstrates a hemorrhage in the right dorsal medulla. Brain MRI (C) and brain artery digital subtraction angiography (D, E) show the arteriovenous fistula at the right craniocervical junction fed by the meningeal branch of the vertebral artery (arrows), draining upwards into the anterior medullary vein (arrowhead) and the superior petrosal sinus subsequently.*

arteriovenous fistula (AVF) in the medulla, which may have caused the bleeding.

## Discussion

Brain stem hemorrhages occur most often in the pons, whereas medullary hemorrhages are very rare.[1] To the best of our knowledge, there is only 1 review concerning medullary hemorrhages; 16 cases of medullary hemorrhage were retrospectively analyzed.[2] We investigated 64 cases who were diagnosed as a brain stem hemorrhage at our hospital from April 2009 to December 2017. Among these, 62 cases were pontine hemorrhage; the other 2 (3%) were medullary hemorrhages (the cases described herein). Most of dural AVFs are considered benign lesions. The annual incidence of hemorrhage at the dural AVFs was only 1.8%.[3] In patients treated conservatively, the fistula could occlude spontaneously.[4] Some cases were diagnosed as medullary AVFs,[5,6] but those clinical courses, especially the occurrence of bleedings, remain unclear. Noncardiac ARDS caused by changes in a patient's intracranial condition are known as causes of NPE.[7] Abnormal sympathetic discharge from "NPE trigger zones" could induce pulmonary venoconstriction, which leads to ARDS.[8] The trigger zones are mainly located in the hypothalamus, as well as the nucleus solitarius and area postrema of the medulla. Direct injury to those areas has been implicated in the generation of isolated neurogenic pulmonary edema.[9] The disruption of the trigger zone reproduced experimental NPE in several animal studies.[9] One case report indicated the possible association between medullary hemorrhage and NPE.[10] A retrospective study of 210 patients with ischemic stroke and transient ischemic attack showed that NPE likely occurred in the setting of brainstem infarction.[11] In addition, Ochiai et al found that patients with a subarachanoid hemorrhage based on the rupture of an aneurysm in a vertebral artery are complicated by NPE more often compared to patients with a subarachanoid hemorrhage based on the rupture of an internal carotid artery or anterior communicating artery.[12] These findings support a strong relationship between a medullary lesion and the development of acute NPE. In conclusion, we experienced 2 rare cases with medullary hemorrhage. AVF can be a cause of the medullary hemorrhage, and NPE could complicate its clinical course.

## Supplementary Materials

Supplementary data to this article can be found online at doi:10.1016/j.jstrokecerebrovasdis.2018.09.027.

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