

Case Report

# Spontaneous subdural hematoma concomitant with subarachnoid hemorrhage and intracerebral hemorrhage due to ruptured brain arteriovenous malformation treated with curative embolization

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## ABSTRACT

**Introduction:** An arteriovenous malformation (AVM) consists of a tangled cluster of dilated blood vessels forming anomalous communication between arterial and venous systems without capillary bridging. The most likely manifestations of a ruptured AVM are intracerebral hemorrhage (ICH), subarachnoid hemorrhage (SAH), or intraventricular hemorrhage (IVH). Subdural hematoma (SDH) is exceptional in cases of ruptured brain arteriovenous malformation (BAVM).

**Case Report:** A 30-year-old female was referred to the Emergency Room with a major complaint of sudden thunderclap headache 1 day before admission. The patient also complained of double vision and left ptosis that lasted for 1 day. There was no other complaint or history of hypertension, diabetes, or trauma. Non-contrast head computed tomography (CT) showed ICH-SAH-SDH on the left side of the brain which was not typical of a hypertensive pattern. The secondary ICH has a score of 6, indicating that 100% of the bleeding might be caused by underlying vascular malformation. Furthermore, cerebral angiography showed cortical plexiform BAVM on the left occipital lobe, and the patient underwent curative embolization.

**Discussion:** Spontaneous SDH is very rare, and there are some hypotheses about the reasons for its occurrence. First, brain movement stretches the arachnoid layer that adheres to the AVM, resulting in direct bleeding into subdural space. Second, high-flow pia-arachnoid ruptured blood might extravasate into subdural space. Finally, the ruptured cortical artery that connects the cortex and dura layer (bridging artery) might also cause SDH. Some scoring systems are useful in BAVM, and this study selected endovascular embolization for the patient.

**Conclusion:** Brain AVM rupture usually causes ICH, SAH, or IVH. Clinicians must be more aware of spontaneous SDH because it might be caused by a vascular malformation, although it is rare.

**Keywords:** Arteriovenous malformation, Embolization, Cerebral hemorrhage, Subarachnoid hemorrhage, Subdural hematoma

## INTRODUCTION

A stroke, also known as cerebrovascular accident, is an acute assault on the perfusion or blood vessels of the brain. It is caused by blockage, such as embolism and thrombosis, or ruptured blood vessels.<sup>[1]</sup> A vascular malformation, such as arteriovenous malformation (AVM) or aneurysm, is one of the causes of hemorrhagic stroke. AVM consists of a tangled cluster of dilated blood vessels forming anomalous communication between arterial and venous systems without capillary bridging.<sup>[2]</sup> The incidence of AVM has been estimated to range between 1.12 and 1.42/100,000 people annually. Its most common presentations include bleeding, seizure, and headache, accounting for 50%, 30%, and 5–14%, respectively.<sup>[3]</sup> However, ruptured AVM most likely manifests as intracerebral hemorrhage (ICH), subarachnoid hemorrhage

(SAH), or intraventricular hemorrhage (IVH).<sup>[2-4]</sup> Subdural hematoma (SDH) is an exception in cases of ruptured brain AVM. There have been only five reported cases so far.<sup>[2,4]</sup>

This study presents a rare case of SDH concomitant with ICH and SAH in brain AVM with an intranidal aneurysm successfully treated with curative embolization. Based on several studies in Indonesia, ruptured aneurysmal SAH has a mortality rate of 20.8–53.1%, higher than other Southeast Asia countries.<sup>[5]</sup> The location of an aneurysm is based on the Circle of Willis' anatomical variations, which is vital to maintain stable cerebral perfusion.<sup>[6]</sup>

## CASE REPORT

A 30-year-old female was referred to the emergency room (ER) with a major complaint of sudden thunderclap headache

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Received: 03 January 2023 Accepted: 15 March 2023 Epub Ahead of Print: 05 April 2023 Published: 03 May 2023 DOI: 10.25259/JNRP\_13\_2023

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a day before admission. This was the worst headache, with a numeric rating scale (NRS) of 10. The patient also complained of double vision and left ptosis that lasted for 1 day. There was no other complaint or history of hypertension, diabetes, or trauma.

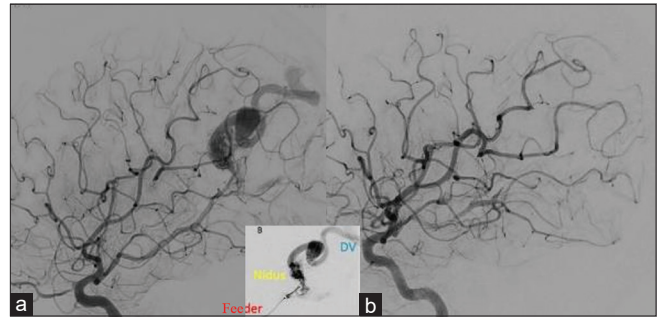
On arrival at the ER, the headache had gotten better with NRS 1–2. Non-contrast head computed tomography (CT) showed ICH, SAH, and SDH on the left side of the brain, which was not consistent with a hypertensive pattern. The secondary ICH had a score of 6, indicating that 100% of the bleeding is caused by underlying vascular malformation. Cerebral angiography showed a plexiform-type cortical AVM with an intranidal aneurysm on the left occipital lobe. The AVM is supplied by a feeder artery from the P4 distal branch of the left posterior cerebral artery and drained by a vein that connects to the occipital cortical vein and superior sagittal sinus. Curative embolization was done 2 days later. First, a microcatheter over microwire was inserted into the feeder artery, and adhesive liquid n-BCA glue (Lipiodol 1:2) with 30% concentration was injected twice, followed by 1:3 with 25% concentration once. The feeder artery was occluded and getting collateral from the pial vessels of the left middle cerebral artery. The procedure was carried out for curative purposes, and complete obliteration of the AVM was achieved [Figure 1]. After 14 days, the second head CT showed a significant reduction and absorption of the bleeding, and the patient was discharged in better condition [Figure 2]. The value of headache NRS ranged from 0 to 1, and there were neither new neurological deficits nor complaints after the procedure.

## DISCUSSION

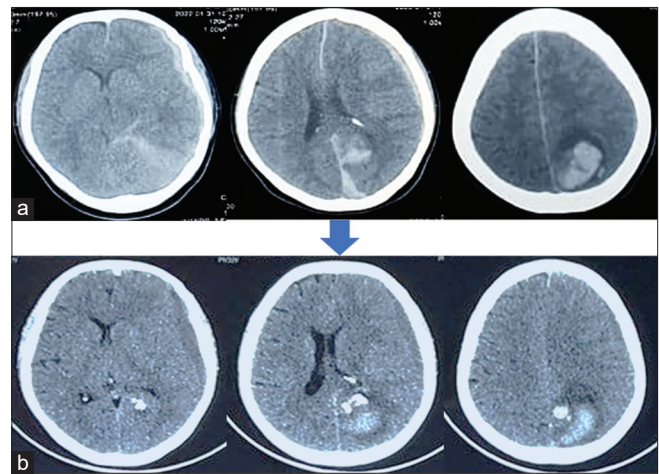
The five subcategories of vascular malformation include cavernous, telangiectasia, varix, venous, and arteriovenous.<sup>[4]</sup> In this study, the patient has symptomatic AVM with bleeding and headache.

Topographically, brain AVM is classified into some subtypes, namely, gyral, sulcal, mixed sulco-gyral, subcortical, and deep AVM. Gyral AVM is usually round-shaped, resides in specific gyrus, and is enveloped by a cortex. On the other hand, huge gyral AVM can expand to the subcortical, even the wall of ventricles. Sulcal AVM is triangular-shaped, with the base pointing to the cortex and the apex to the ventricle. Feeder arteries come from branches of the pial artery, also by perforating artery when located in the periventricular.<sup>[7]</sup> The AVM of this study was mixed-type, and it does not have a direct connection with the dura-arachnoid layer. Therefore, the most common manifestations are ICH, SAH, and IVH.<sup>[2-4]</sup> Another bleeding risk factor is the routine use of anticoagulants and antiplatelet agents, but there was no consumption history of these drugs in the patient.<sup>[8]</sup>

Spontaneous SDH occurs with AVM and intranidal aneurysms, and there are some hypotheses about the



**Figure 1:** (a) Initial cerebral angiography versus and (b) post-curative embolization.



**Figure 2:** (a) Initial non-contrast head computed tomography and (b) 14 days after onset (post-curative embolization).

reasons for its occurrence. First, brain movement causes stretching of the arachnoid layer that adheres to the AVM and causes direct bleeding into subdural space. Second, high-flow pia-arachnoid ruptured blood might extravasate into subdural space. Finally, the ruptured cortical artery that connects the cortex and dura layer (bridging artery) might also cause SDH.<sup>[4]</sup> Intranidal aneurysms have a thin wall and are closer to the terminal feeder, such that it becomes more susceptible to the increase of intra-arterial pressure and rupture. This study recommends CT angiography as a screening tool for spontaneous SDH. Furthermore, contrast extravasation from cortical vessels can help localized bleeding focus and detect a cortical AVM.<sup>[7]</sup>

The available treatment options for AVM include excision surgery, stereotactic radiosurgery, endovascular embolization, or conservative. There are certain scoring systems that are useful in patients with AVM. The Spetzler-Martin grading system can be used to assess the risk and difficulty of surgery. In comparison, AVM Embocure Score (AVMES) and Feliciano grading are useful for predicting the embolization treatment outcome.<sup>[7]</sup> In this study, the patient's AVMES was 3, indicating 100% complete obliteration without

complication. Moreover, the Feliciano grade was 1, indicating that embolization can achieve complete obliteration. Based on this consideration, endovascular embolization was chosen.

## CONCLUSION

SDH is extremely rare as a result of brain AVM rupture, and it usually causes ICH, SAH, or IVH. It is believed that the adhesion of the AVM vessel to the arachnoid membrane and its strain could lead to SDH. Clinicians must be more aware of spontaneous SDH without a history of trauma, because it might be caused by a vascular malformation, although it is rare. Furthermore, proper scores can be used for the treatment option to predict the best outcome.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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**How to cite this article:** Soedjono HK, Sani AF, Ekowahono J, Kurniawan D. Spontaneous subdural hematoma concomitant with subarachnoid hemorrhage and intracerebral hemorrhage due to ruptured brain arteriovenous malformation treated with curative embolization. *J Neurosci Rural Pract* 2023;14:368-70.