

# CASE REPORT

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## A rare pure intraventricular hemorrhage caused by ruptured internal carotid artery-anterior choroidal artery aneurysm: a case report and literature review

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### ABSTRACT

Although intraventricular hemorrhage (IVH) frequently develops secondary to intraparenchymal or subarachnoid hemorrhage, pure IVH—non-traumatic spontaneous intracranial hemorrhage confined to the cerebral ventricular system—is rare. Moreover, pure IVH caused by ruptured proximal aneurysm is an extremely rare but life-threatening condition. Herein, we present a case of pure IVH due to a ruptured internal carotid artery -anterior choroidal artery (ICA-AChA) aneurysm and review related literature. A 77-year-old man presenting with altered mental status was hospitalized due to a massive pure IVH with ventriculomegaly. The patient was conservatively managed because his consciousness level improved. On the following day, computed tomography angiography revealed a right ICA-AChA aneurysm embedded in the temporal lobe adjacent to the anterior part of the inferior horn of the lateral ventricle, which was consistent with the hemorrhagic origin. Coil embolization of the aneurysm was successfully performed, and the postprocedural course was uneventful. To date, only a few cases have described pure IVH caused by a ruptured proximal aneurysm. If the ruptured proximal aneurysm remains unnoticed, catastrophic rehemorrhage may occur. Aneurysm obliteration should precede the treatment of acute hydrocephalus. Neurosurgeons/neurointerventionalists must be aware about pure IVH caused by a ruptured proximal aneurysm even if it rarely occurs.

Keywords: acute hydrocephalus, isolated intraventricular hemorrhage, primary intraventricular hemorrhage, ruptured cerebral aneurysm, solitary intraventricular hemorrhage

#### Abbreviations:

AChA: anterior choroidal artery

ICA: internal carotid artery

IVH: intraventricular hemorrhage

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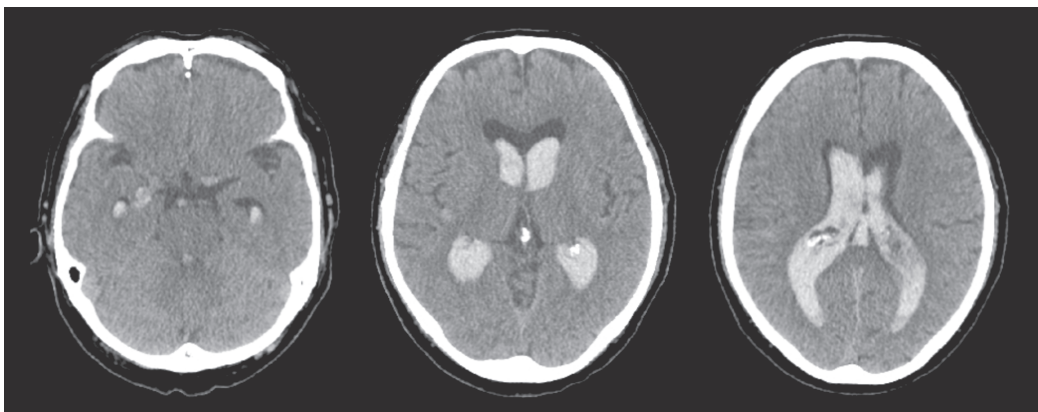
## INTRODUCTION

Ruptured cerebral aneurysm is life-threatening and requires urgent medical attention. It commonly causes subarachnoid hemorrhage (SAH), which is occasionally associated with intraparenchymal hemorrhage (IPH), intraventricular hemorrhage (IVH), or subdural hematoma. However, extremely rare cases of pure IVH (isolated, primary, or solitary IVH), that is, non-traumatic spontaneous intracranial hemorrhage in the cerebral ventricular system without any evidence of IPH or SAH that is caused by ruptured proximal aneurysms such as supraclinoid internal carotid artery (ICA), anterior communicating artery (ACoA), and basilar artery aneurysms, have been reported.<sup>1-8</sup> In such cases, the ruptured cerebral aneurysm can be overlooked, potentially leading to fatal rebleeding. Herein, we present a rare case of pure IVH due to a ruptured ICA-anterior choroidal artery (AChA) aneurysm and a review of related literature.

## CASE REPORT

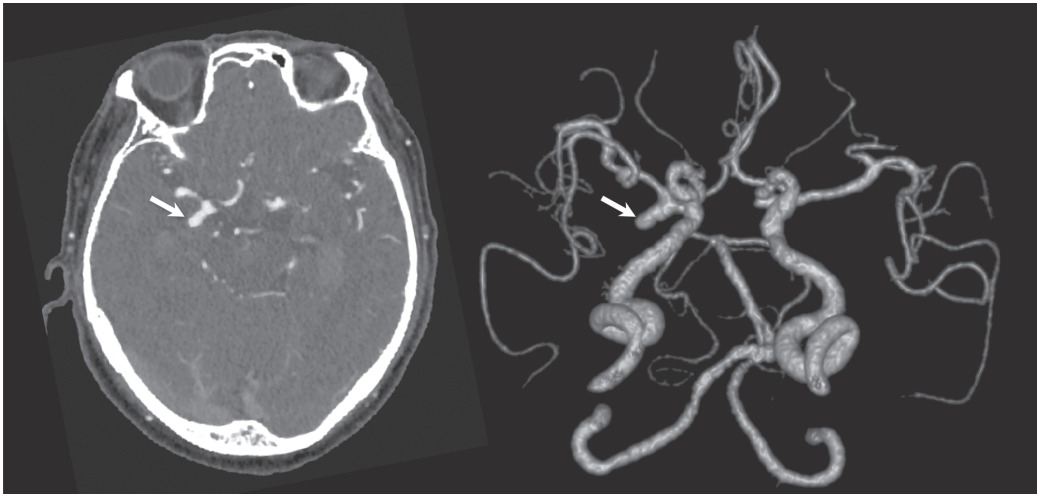
A 77-year-old man with altered mental status (Glasgow Coma Scale [GCS] score, E2V2M5) presented to a nearby hospital. He had no history of hypertension, dyslipidemia, diabetes mellitus, stroke, and smoking or alcohol abuse. On admission (day 0), computed tomography (CT) revealed a massive pure IVH with ventriculomegaly but no SAH (Fig. 1). Owing to the patient's gradually improving consciousness (GCS score, E3V4M6), conservative treatment was applied.

On day 1, CT angiography (CTA) disclosed a right ICA-AChA aneurysm (11 mm) embedded in the temporal lobe contiguous to the anterior part of the inferior horn of the lateral ventricle (Fig. 2). The hematoma distribution indicated that the ICA-AChA aneurysm was compatible with the hemorrhagic origin. On the same day, the patient was transferred to our institution for emergent endovascular treatment. Under general anesthesia, a 6F Simmons guiding sheath (6F Axcelguide Stiff-J-1, 90 cm; Medikit, Tokyo, Japan) was introduced into the right common carotid artery via a right transradial approach.<sup>9-12</sup> After a 6F intermediate catheter (6F SOFIA, 115 cm; MicroVention-Terumo, Aliso Viejo, California, USA) was delivered into the ICA petrous segment, coil embolization of the aneurysm was performed using the double-catheter technique. Aneurysm obliteration and AChA patency were confirmed on postprocedural angiography (Fig. 3).

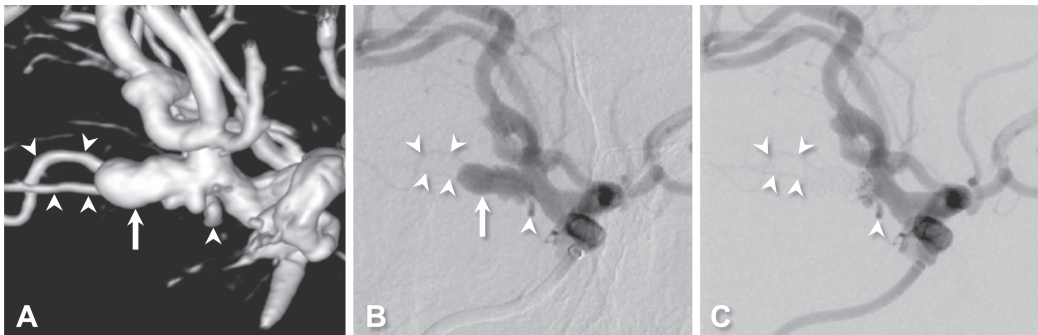


**Fig. 1** Head computed tomography (CT) images on admission

CT shows a massive pure intraventricular hemorrhage with ventriculomegaly but no evidence of subarachnoid hemorrhage.



**Fig. 2** Preprocedural computed tomography angiography (CTA) source image (left) and three-dimensional CTA image (right). The CTA source image demonstrates a right internal carotid artery-anterior choroidal artery (ICA-AChA) aneurysm (arrows) compatible with a hemorrhagic origin. The ICA-AChA aneurysm projected posterolaterally and is embedded in the temporal lobe adjacent to the anterior part of the inferior horn of the lateral ventricle.



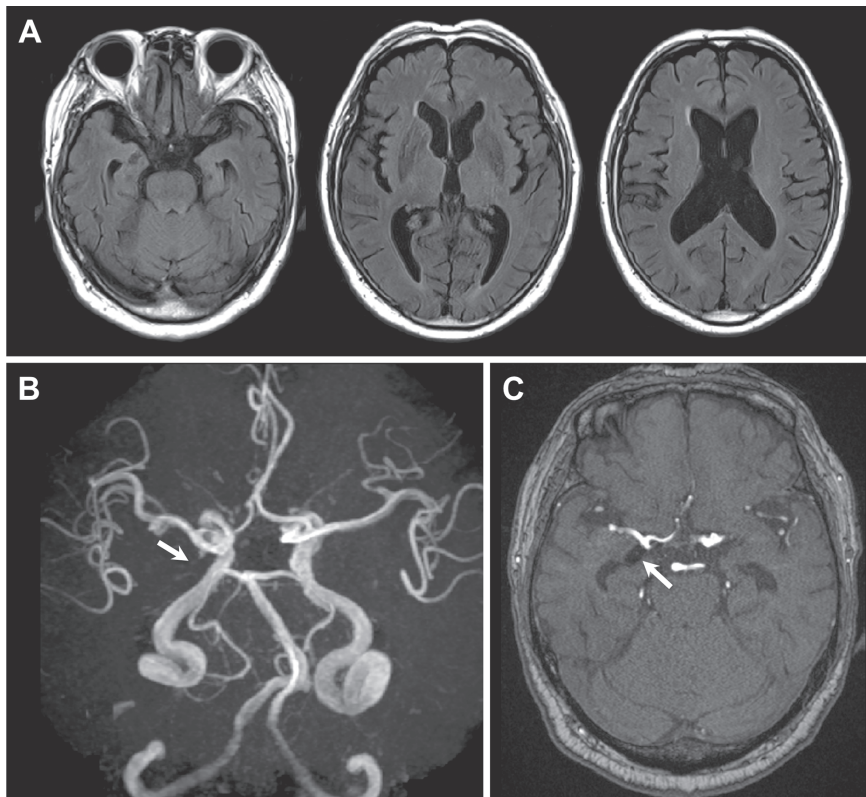
**Fig. 3** Preprocedural three-dimensional rotational angiography (3DRA) (A)/angiography (B) and postprocedural angiography images (C)

**Fig. 3A:** Right anterior oblique view of the preprocedural 3DRA image showing a right internal carotid artery-anterior choroidal artery (ICA-AChA) aneurysm (arrow). The right AChA (arrowheads) originates from the aneurysm neck.

**Fig. 3B:** Right anterior oblique view of the preprocedural angiography image showing the right ICA-AChA aneurysm (arrow) and right AChA (arrowheads).

**Fig. 3C:** Right anterior oblique view of the postprocedural angiography image confirming the successful obliteration of the ICA-AChA aneurysm and right AChA patency (arrowheads).

The postprocedural course was uneventful. The patient's consciousness gradually recovered. On day 11 and day 18, follow-up magnetic resonance angiography revealed no evidence of cerebral infarction and delayed cerebral vasospasm. On day 21, satisfactory aneurysm obliteration was confirmed on follow-up angiography. On day 36, magnetic resonance imaging revealed that the IVH was completely washed out without ventriculomegaly (Fig. 4). On day 39, the patient was discharged home, with clear consciousness.



**Fig. 4** Postprocedural fluid-attenuated inversion recovery (FLAIR) (A) and magnetic resonance angiography (MRA) maximum intensity projection (MIP) images (B) and MRA source image (C) 37 days after onset of hemorrhage

**Fig. 4A:** FLAIR images showing the intraventricular hemorrhage completely washed out with no evidence of ischemic stroke and ventriculomegaly.

**Fig. 4B, 4C:** Follow-up MRA MIP and source images showing no evidence of recanalization of the treated aneurysm (arrows).

## DISCUSSION

IVH frequently develops secondary to IPH or SAH. However, pure IVH is rare, accounting for 0.31% and  $\leq 3\%$  of all stroke and intracranial hemorrhage cases.<sup>2,5</sup> Initial manifestations include headache (69%), altered mental status (66%), and nausea/vomiting (53%).<sup>2</sup> The overall in-hospital mortality was 39%. High patient age, low initial GCS score, large amount of IVH, and acute hydrocephalus are poor independent prognostic predictors.<sup>2,5</sup> While Moyamoya disease, dural arteriovenous fistulas, intraventricular/paraventricular cavernous malformations, or intraventricular/paraventricular tumors rarely cause pure IVH, cerebral aneurysms and arteriovenous malformations are the most common vascular causes.<sup>2,13</sup> Pure IVH caused by ruptured cerebral aneurysm has high morbidity and mortality.<sup>6</sup> The aneurysms were reported to be predominantly peripheral, including intraventricular aneurysms (eg, distal AChA aneurysm, medial posterior choroidal artery aneurysm, lateral posterior choroidal artery aneurysm, lenticulostriate artery aneurysm, distal posterior inferior cerebellar artery aneurysm and distal superior cerebellar artery aneurysm) which usually are very small and occur secondary to arteriovenous malformation, Moyamoya disease,

**Table 1** Summary of case reports of pure IVH caused by ruptured proximal aneurysms

| Age (years)/sex                            | Initial manifestation                           | Acute hydrocephalus | Ruptured aneurysm |           |           |              | Cerebral vasospasm | Treatment                | Discharge GOS score |
|--|---|---------------------|-------------------|-----------|-----------|--------------|--------------------|--------------------------|---------------------|
|  |   |                     | Modality          | Diagnosis | Size (mm) | Location     |                    |                          |                     |
| Thai et al. <sup>1</sup> (2005)            | Altered mental status (coma)                    | Yes                 | Angiography       | 1         | 24        | BA tip       | NA                 | Coiling                  | 2                   |
| Flint et al. <sup>2</sup> (2008)           | NA  | NA                  | Angiography       | NA        | NA        | Pericallosal | NA                 | NA                       | NA                  |
| Prasad et al. <sup>3</sup> (2009)          | Headache/vomiting                               | No                  | CTA               | 1         | NA        | ICA-PCoA     | Lateral            | Coiling                  | 5                   |
| Nakagawa et al. <sup>4</sup> (2014)        | Headache/altered mental status (confusion)      | Yes                 | CTA               | 1         | NA        | ICA-PCoA     | Lateral            | Coiling followed by EVD  | 5                   |
| Lee et al. <sup>5</sup> (2017)             | NA  | NA                  | NA                | NA        | NA        | ACoA         | NA                 | NA                       | NA                  |
| Anagnostopoulos et al. <sup>6</sup> (2019) | Altered mental status (coma)                    | Yes                 | Angiography       | 3         | NA        | ICA-PCoA     | Postero-lateral    | EVD followed by coiling  | 5                   |
| Kim et al. <sup>7</sup> (2019)             | Headache/altered mental status (disorientation) | Yes                 | CTA               | 1         | 7         | ICA-AChA     | Postero-lateral    | Coiling followed by LD   | 4                   |
| Oberman et al. <sup>8</sup> (2020)         | Headache/nausea                                 | Yes                 | CTA               | 1         | 7         | ACoA         | Antero-superior    | Clipping followed by EVD | 1                   |
| Present case                               | Altered mental status (stupor)                  | Yes                 | CTA               | 2         | 11        | ICA-AChA     | Postero-lateral    | Coiling                  | 5                   |

AChA: anterior choroidal artery

ACoA: anterior communicating artery

BA: basilar artery

CTA: computed tomography angiography

EVD: external ventricular drainage

F: female

GOS: Glasgow Outcome Scale

ICA: internal carotid artery

IVH: intraventricular hemorrhage

LD: lumbar drainage

M: male

NA: not available

PAD: post-admission day

PCoA: posterior communicating artery

infection, or head trauma.<sup>2,14,15</sup> As they occasionally disappear spontaneously, conservative treatment may be used for ruptured intraventricular aneurysms if there is no possible treatment.<sup>14</sup> By contrast, pure IVH caused by ruptured proximal aneurysms is extremely rare, with only 9 cases, including our case, reported so far (Table 1).<sup>1-8</sup> Such aneurysms have a rebleeding risk, which can be potentially devastating and fatal. The case reported by Oberman et al was diagnosed as pure IVH caused by a ruptured ACoA aneurysm on day 0.<sup>8</sup> On day 2, the patient's consciousness deteriorated before treatment, which indicated rebleeding of the aneurysm. The patient died despite aneurysm clipping and external ventricular drainage. To prevent catastrophic rebleeding, the ruptured cerebral aneurysm must be correctly identified and treated immediately. The reported aneurysms were  $\geq 7$  mm (range, 7–24 mm) and located in the ICA-posterior communicating artery (PCoA; n = 3), ICA-AChA (n = 2), ACoA (n = 2), pericallosal artery (n = 1), and basilar artery (n = 1; Table 1). Pure IVH can occur theoretically if the aneurysm protrudes into the cerebral ventricular system. In clinical practice, all ICA aneurysms (n = 5) were embedded in the temporal lobe adjacent to the anterior part of the inferior horn of the lateral ventricle (Table 1). Diagnosis was performed using angiography (n = 3) and CTA (n = 5; Table 1). Angiography is a promising modality for identifying potentially treatable causes of pure IVH, including very small intraventricular aneurysms.<sup>2</sup> Considering aneurysm size ( $\geq 7$  mm), CTA will be a useful diagnostic modality for pure IVH caused by ruptured proximal aneurysm because it can detect the anatomical relationships between the aneurysm, cerebral ventricular system, and hematoma distribution. Acute hydrocephalus occurred in 6 cases and was treated conservatively (n = 2), with coiling/clipping followed by cerebrospinal fluid drainage (n = 3), and with cerebrospinal fluid drainage followed by coiling (n = 1; Table 1). In another reported case, external ventricular drainage was used for acute hydrocephalus associated with pure IVH before identification of the causative vascular lesion (day 0).<sup>6</sup> Fortunately, aneurysm coiling was achieved without rebleeding after detection of the ruptured ICA-PCoA aneurysm on day 2. For intracranial pressure control and/or hematoma removal, external ventricular drainage, and/or endoscopic surgery should be considered urgently for acute hydrocephalus associated with IVH, especially in patients with decreased consciousness,<sup>2,16,17</sup> but these could cause rebleeding from the ruptured aneurysm,<sup>18</sup> which can be a pitfall in patients with pure IVH caused by ruptured proximal aneurysms. In patients with pure IVH, vascular imaging is mandatory, not optional. Surgical/endovascular treatment for ruptured aneurysm should precede treatment for acute hydrocephalus. Neurosurgeons/neurointerventionalists must be knowledgeable about pure IVH caused by a ruptured proximal aneurysm even if it rarely occurs.

#### AUTHOR CONTRIBUTION

YS and YH contributed equally as first authors to this work.

#### CONFLICTS OF INTEREST

The authors have no conflicts of interest to disclose.

#### ETHICS APPROVAL

This study was performed in accordance with the ethical standards of the 1964 Declaration of Helsinki and its later amendments.



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