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**ABOUT COVER**

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## Hemorrhagic transformation of ischemic cerebral proliferative angiopathy: A case report

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### Abstract

#### BACKGROUND

Cerebral proliferative angiopathy (CPA) is a rare vascular disease characterized by the presence of diffuse vascular proliferation, progressive vascular hyperflow and vasodilation of multiple vessels in the normal brain parenchyma. Unlike cerebral arteriovenous malformations, CPA has a mixed appearance between that of lesions with cell proliferation and endothelial proliferation. To date, the pathogenesis of CPA is unclear, in which changes induced by cortical ischemia in the elastic layer of the blood supply artery and smooth muscle cells may be involved.

#### CASE SUMMARY

In this article, we retrospectively analyzed a case of hemorrhagic transformation of ischemic CPA diagnosed by digital subtraction angiography and reviewed the related literature for further exploration of its pathogenesis, diagnosis and treatment.

#### CONCLUSION

The information in the present case report may facilitate further clinical research on this cerebrovascular disease.

**Key Words:** Hemorrhagic transformation; Cerebral proliferative angiopathy; Pathogenesis; Diagnosis; Treatment; Case report

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**Core Tip:** This article reports a very rare case of ischemic cerebrovascular disease, namely ischemic cerebral proliferative angiopathy. After consulting major databases, we found that cases of ischemic and hemorrhagic cerebral proliferative angiopathy

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have been reported, but the biggest feature of this case is the phenomenon of hemorrhagic transformation after ischemia. So far, there are few reports of such diseases globally, and their pathogenesis is currently unclear. The diagnosis and treatment methods are limited. Therefore, this article reviews relevant literature to better understand this rare disease and provides clues for further clinical research.

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## INTRODUCTION

Cerebral proliferative angiopathy (CPA) is defined as an atypically chronic ischemic cerebrovascular disease that appears to be significantly different from normal arteriovenous malformation (AVM) in epidemiology, clinical manifestations, angiographic and histopathological features, natural history and management[1]. In CPA, a large and sparse lesion caused by progressive angiogenesis can be seen in the normal brain parenchyma. Ischemia induced by hypoperfusion around the lesion is considered to lead to that excessive proliferation[2]. CPA is less common than AVM, and hemorrhagic transformation of ischemic CPA is much rarer.

## CASE PRESENTATION

### Chief complaints

Our patient was a 76-year-old male with dizziness, headache and left hemiparesis and numbness.

### History of present illness

The patient was diagnosed with hemorrhagic cerebral infarction and was treated with mannitol and glycerol fructose to reduce intracranial pressure.

### History of past illness

The patient had a history of hypertension, and no oral medications were applied.

### Personal and family history

The patient smoked for decades and presently quit smoking. In addition, he occasionally drank alcohol. No family hereditary diseases and tumors were reported.

### Physical examination

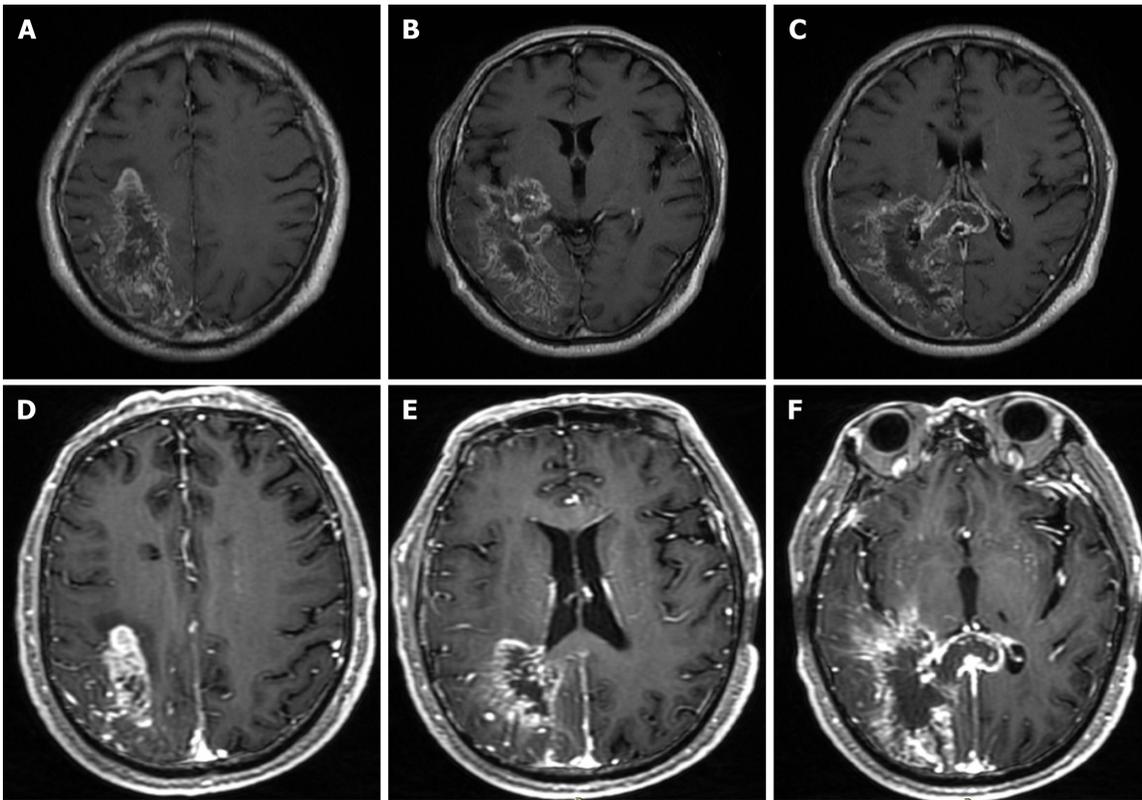
At the time of admission, the main clinical positive signs of this patient were as follows: positive horizontal nystagmus; both eyes were blurred to the left; the left limb muscle strength was grade IV; the left limbs' shallow sensations were decreasing; and the left pathological sign was positive.

### Laboratory examinations

Cerebrospinal fluid routine, biochemistry and serum paraneoplastic neurological syndrome test results were negative. Additionally, there were no significant abnormalities in blood examinations.

### Imaging examinations

A magnetic resonance imaging (MRI) scan was performed, which revealed a massive cerebral infarction in the right temporal lobe, parietal lobe, occipital lobe and the corpus callosum accompanied by minor bleeding and gliosis within. In addition, brain enhanced MRI showed irregular ring enhancement in the right temporo-occipital junction area and corpus callosum compression, and the posterior horn of the right lateral ventricle was compressed (Figure 1). Brain computed tomography angiography



**Figure 1 Brain magnetic resonance imaging.** T1-weighted images showed different locations of a massive cerebral infarction accompanied by minor bleeding and gliosis. A: The right parietal lobe; B: The right temporal lobe and the occipital lobe; C: The right temporal lobe, the occipital lobe and the corpus callosum. Enhanced T1-weighted images showed irregular ring enhancement; D: The right parietal lobe; E: the right temporal lobe and the occipital lobe; F: The right temporal lobe, the occipital lobe and the corpus callosum.

revealed intracranial artery stenosis and hyperplasia (Figure 2).

Chest and abdominal pelvic computed tomography and brain magnetic resonance venography. Digital subtraction angiography showed a large number of proliferative blood vessels in the blood supply area of the right middle cerebral artery. A small number of proliferative blood vessels were also seen in the A4 segment of the right anterior cerebral artery. A large number of proliferative blood vessels were visible in the P3 or P4 segment of the posterior artery (Figure 3).

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## FINAL DIAGNOSIS

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Hemorrhagic CPA after ischemia

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## TREATMENT

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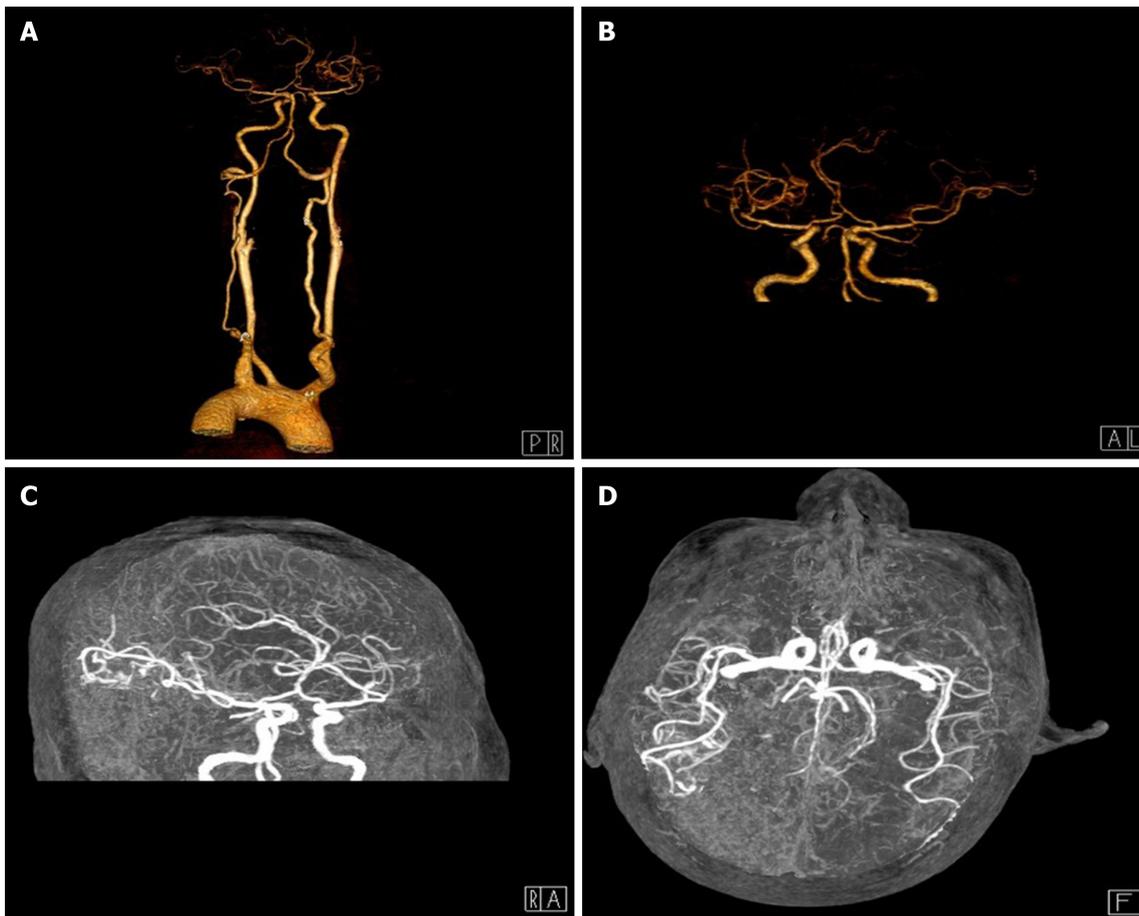
In this case, symptomatic treatments such as lowering intracranial pressure and fluid supplementation were the main treatments. Considering that the risk of surgical treatment was too high, the family refused.

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## OUTCOME AND FOLLOW-UP

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At his 1-year telephone follow-up, we learned that the aforementioned clinical symptoms had been progressively worsening with a disturbance of consciousness, and the patient had eventually passed away.



**Figure 2 Brain computed tomography angiography.** Computed tomography angiography showed different locations of artery stenosis and hyperplasia. A: Mild stenosis in the internal carotid arteries on both sides at the beginning; B: The right middle cerebral artery with hyperplasia; C: The thickness of the anterior cerebral arteries on both sides was uneven; D: The posterior cerebral artery on the right was slender.

## DISCUSSION

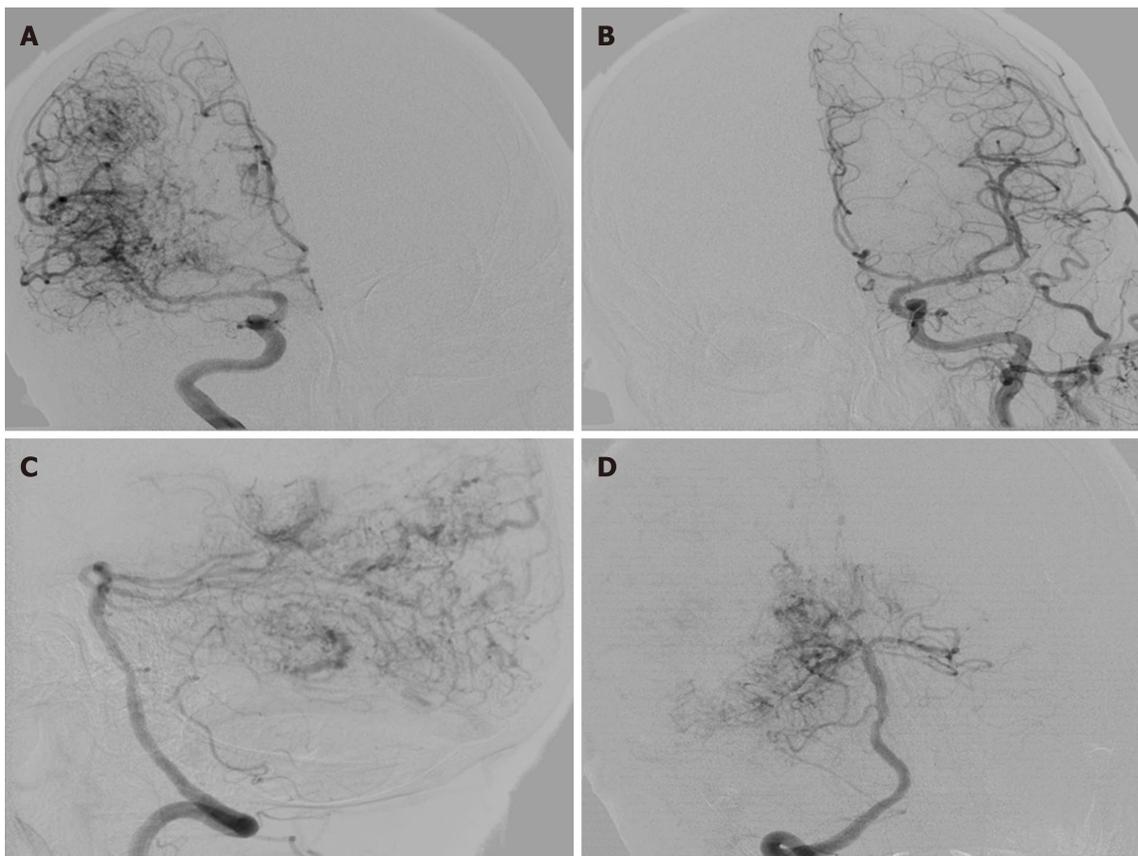
Recently, Lasjaunias *et al*[1] retrospectively analyzed 1434 AVM databases, where they identified CPA in 49 cases with an age range of 10-65 years, with the highest proportion in young women. In addition, Puerta *et al*[3] reported an 8-year-old child suffering from CPA. However, our patient was a 76-year-old male. Other investigations[4-6] showed that CPA accounted for approximately 2%-4% of all AVMs, indicating that the right cerebral hemisphere was the most common location, especially the temporal lobe. CPA usually manifested as epilepsy, headache and progressive neurological deficit (predominantly ischemia). Our patient had lesions in the right temporal, parietal and occipital lobes. The most important finding was that in the case described here, the patient had postischemic hemorrhagic transformation. To date, this transformation phenomenon has not been largely reported in the literature, and thus we collected and summarized the information available on all cases of hemorrhagic CPA to establish its clinical features[7-13] (Table 1).

CPA was diagnosed by its typical vascular imaging features. The major manifestation of CPA on MRI was diffuse lesions with a densely enhanced vascular network structure mixed with normal brain parenchyma. In some CPA patients, the drainage veins were reduced, with varying degrees of vein thickening on MRI. On digital subtraction angiography, the blood vessels were in a muddy shape with a scattered distribution, manifested in a small shunt volume, lack of dominant arterial blood supply period, delayed arterial phase and early venous phase. The patient had a large number of proliferative blood vessels in the blood supply area of the right middle cerebral artery. Proliferated blood vessels were also visible in the A4 segment of the right anterior cerebral artery. No obvious abnormalities were detected in the left middle cerebral artery and the anterior cerebral artery. The vertebral artery angiography showed extensive hyperplasia in the P3 or P4 segment of the right posterior cerebral artery. The draining veins were clearly visible, and no abnormality was found in the left posterior cerebral.

**Table 1** Review of all hemorrhagic cerebral proliferative angiopathy published to date

Ref.	Age/sex	Clinical presentation	Location	Management	Follow-up
Maekawa <i>et al</i> [7], 2012	62/F	Gait instability and dysarthria	Tectum and cerebellar vermis	Symptomatic	9 yr; died
Kumar <i>et al</i> [8], 2015	66/M	Headache	Left cerebellar hemisphere	Asymptomatic	12 mo; no deficits
Bilaj <i>et al</i> [9], 2016	24/M	Headache and seizures	Left temporal lobe	Frontotemporal craniotomy	NA
Kimiwada <i>et al</i> [10], 2019	13/M	Headaches and reversible focal neurological deficits	Left frontal and parietal lobes	Indirect revascularization procedure	2 yr; stable symptoms
Maekawa <i>et al</i> [11], 2018	12/F	Intractable headaches	Left cerebellar hemisphere	Symptomatic	30 yr; recurrence of cerebral hemorrhage
Giragani <i>et al</i> [12], 2018	12/M	Headache	Cerebellar hemisphere and cerebellar vermis	Asymptomatic	6 mo; no deficits
Beniwal <i>et al</i> [13], 2020	12/M	Trivial trauma and altered sensorium with hemorrhage in imaging	Right cerebellar hemisphere	Symptomatic	18 mo; no deficits
Current case	76/M	Dizziness, headache and left hemiparesis and numbness	Right cerebellar hemisphere	Symptomatic	12 mo; died

F: Female; M: Male; NA: Not available.



**Figure 3** Brain digital subtraction angiography. Anteroposterior view of a right internal carotid artery (ICA) angiogram. A: Anteroposterior view of a left ICA angiogram; B: Lateral view of a right posterior cerebral artery (PCA) angiogram; C: Anteroposterior view of a right PCA angiogram; D: Diffuse vascular malformation can be seen in the right cerebral hemisphere.

CPA may be cured surgically by embolization and using stereotactic radiotherapy, which could increase the blood flow to the hypoperfused brain tissue and prevent future ischemia[14,15]. Considering that the aforementioned treatment methods are risky with poor long-term prognosis, conservative treatment is still the mainstay[16].

In our case, the most characteristic feature of CPA that distinguished it from cerebral AVMs was the presence of recognizable neurons and normal brain tissue intertwined with abnormal blood vessels[17]. The vascular lesions of CPA were unstable, and their continuous growth was detected by neuroimaging at multiple time points. The lesions gradually expanded, accompanied by aggravated clinical symptoms, which indicated that CPA was progressive. After the patient was discharged from the hospital on September 3, 2019, his dysfunction of the limbs worsened again. However, due to the unstable vital signs of the patient, his digital subtraction angiography images were not reviewed. Larger clinical trials are required for a further exploration of the pathogenesis of this disease aimed at the discovery of new therapeutic options.

## CONCLUSION

CPA is a rare ischemic vascular disease, and hemorrhagic transformation after ischemia has been seldom reported. In recent years, increased research attention has been paid to this disease. The pathogenesis, clinical manifestations, diagnosis and treatment of CPA have been initially established. However, in the future, more high-quality clinical research is to be conducted to further explore its pathogenesis, improve therapeutic schedules for minimizing its disability and reduce mortality.

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