A vertebral artery aneurysm that elicited occipital pain, showed no abnormalities on imaging studies, but ruptured the next day: illustrative case

Masaaki Kawauchi, MD,1 Kyongsong Kim, MD, PhD,2 and Go Inokuchi, MD, PhD3

1Department of Neurosurgery, Chiba Shintoshi Rurban Clinic, Inzai, Chiba, Japan; 2Department of Neurological Surgery, Chiba Hokuso Hospital, Nippon Medical School, Inzai, Chiba, Japan; and 3Department of Legal Medicine, Graduate School of Medicine, Chiba University, Chiba, Chiba, Japan

BACKGROUND The authors report the case of a patient with occipital headache whose imaging studies revealed no abnormalities but who died 1 day later due to vertebral artery (VA) aneurysm rupture.

OBSERVATIONS A male in his 40s with no relevant medical history had been taking over-the-counter medication for headache several times a month. One day before he visited our neurosurgery department, he experienced occipital headache, took the usual medicine, and applied a warm compress. Brain magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) studies returned no abnormal findings, and he went home. On the following day, his wife found his lifeless body in rigor mortis and requested a medicolegal autopsy. Preautopsy brain computed tomography showed diffuse subarachnoid hemorrhage (SAH). Histopathologically, there was no obvious VA dissection. The vascular wall at the rupture site lacked internal elastic lamina and media, it was covered only with thin fibrous connective tissue, and the adventitia was expanded. The cause of death was determined to be SAH due to rupture of a VA blister aneurysm.

LESSONS In our patient, brain MRI and MRA studies returned no abnormal findings. However, he died suddenly the next day. Autopsy identified SAH due to rupture of a blister-like VA aneurysm without dissection.

https://thejns.org/doi/abs/10.3171/CASE23723

KEYWORDS vertebral artery; autopsy; diagnosis; MRI; MRA
it was intense in the right posterior neck and aggravated by bathing in hot water and by bending forward. He took the usual over-the-counter medication and applied a warm compress. Although the intensity of the headache was not strong, it was different from the usual pain; it was not a sudden severe pain (thunderclap pain). The next morning, he underwent brain MRI and MRA studies at our neurosurgery department. As shown in Fig. 1, there were no abnormal findings. The diagnosis was migraine, and we prescribed triptans. Although the headache continued, he returned home and had supper with his wife.

Around 1:00 PM the next day, his wife sent an email to him, but it was not read. When she returned home at 7:30 PM, she found his corpse in the living room. Cardiopulmonary arrest and rigor mortis were confirmed, and a medicolegal autopsy was performed a week later.

Preautopsy brain computed tomography (CT) showed diffuse SAH (Fig. 2). At autopsy, catheters were inserted into the VA and ICA for computed tomography angiography (CTA). Leakage of contrast medium from the right VA was observed at a dilated VA portion without branches, and a blood blister–like aneurysm was identified (Figs. 3–5).

FIG. 1. Fluid-attenuated inversion recovery MRI (A–D) and MRA (E) performed 1 day after occipital headache onset. Neither SAH (A–D) nor obvious dilation or constriction (E) of the VA is observed, although the V4 segment is tortuous.

FIG. 2. Preautopsy brain CT showing diffuse SAH.

FIG. 3. Cranial CTA performed at autopsy. Note dilation of the right VA and leakage of contrast medium at the dilation site (arrow).
Histopathology revealed no obvious VA dissection. The vascular wall at the rupture site lacked internal elastic lamina and media and was covered only with thin fibrous connective tissue, and the adventitia was expanded. Inflammatory cells, mainly neutrophils, infiltrated the surrounding area. The cause of death was determined to be SAH due to rupture of a VA blister aneurysm.

Patient Informed Consent

The necessary patient informed consent was obtained in this study.

Discussion

Observations

Based on pathological findings, Mizutani et al.\(^6\) classified 85 nonatherosclerotic cerebral fusiform and dissecting aneurysms in unbranched areas into four types. Type 1 are classic dissecting aneurysms, type 2 show segmental ectasis with intimal thickening, type 3 are dolichoectasic dissecting aneurysms, and type 4 (anterior circulation: \(n = 3\), VA: \(n = 1\)) were saccular aneurysms arising from the arterial trunk. In type 4 aneurysms, the internal elastic layer of the ruptured cerebral aneurysm wall is missing, and the adventitia is expanded and fragile; dorsal ICA wall and blister-like aneurysms might also be included as type 4. Some type 4 aneurysms rebleed easily because of the fragile aneurysmal wall. Three patients with type 4 aneurysms, including one with VA involvement, could not be operated on and died; the fourth patient survived after aneurysm clipping.\(^6\) The blister-like VA aneurysm in our patient was probably type 4 because its wall lacked the internal elastic lamina and tunica media and no intima was observed.

In 40% of autopsies confirming SAH due to cerebral aneurysm rupture, prodromal symptoms were reported; 32% of these patients had a ruptured aneurysm, 2% had a dissecting cerebral aneurysm, and headache was the most common prodromal symptom.\(^1\) In a

FIG. 4. A photograph of the brain reveals widespread SAH at the basal cistern. Note the attached hematoma. Part of the artery is dilated in the enlarged brainstem (yellow arrow).

FIG. 5. A and B: Formalin-fixed right VA. C-F: Pathological findings (EvG staining). A thrombus is seen at the ruptured part of the blood vessel. The vascular wall lacks internal elastic lamina. The media is covered only with thin fibrous connective tissue. There is no artery dissection. The internal elastic lamina is partially torn from ruptured blood vessels. Neutrophils infiltrate the adventitia side of the vascular wall around the ruptured area.
study of 206 patients with ICA dissection, most involved the VA; headache was the most frequent prodromal symptom in patients with or without aneurysm rupture.7

Among 96.4% of patients with intracranial artery dissection, the time from headache onset to SAH was less than 3 days; the average time from headache to an imaging diagnosis was 9.8 days.8 Although such information on patients with blister aneurysms is lacking, the antemortem headache of our patient appears to have been a precursor to the rupture of a new VA blister aneurysm. Brain MRA performed 16 hours after the onset of his occipital pain returned no abnormal findings, although he developed SAH 12 hours later and died.

The results of vessel wall imaging of ruptured cerebral aneurysms have been reported.9–11 Pathological findings on dissecting cerebral aneurysms suggest that gadolinium (Gd)-enhanced MRI studies of the vessel wall can monitor inflammatory processes.10 Circumferential arterial wall enhancement was useful for recognizing ruptured cerebral aneurysms.11 In a prospective study of 145 unruptured cerebral aneurysms, 8 had morphological and circumferential arterial wall enhancement.9 Gd-enhanced MRI of the vessel wall may be useful to evaluate the risk of pathological changes and rupture of the vascular wall,10,11 even when the aneurysm, as in our patient, is not saccular. We are in the process of investigating this issue.

Pathology suggested changes in the vascular wall at the inception of his occipital pain, but no morphological changes were observed on MRI scans; 24 hours after our imaging studies, the aneurysm suddenly expanded, ruptured into a blister-like aneurysm, followed by a fatal SAH. Diagnosticians must be cognizant of the existence of such rare cases.

Because a frequent prodromal symptom elicited by vertebral cerebral aneurysms is occipital or neck pain,1 patients with these symptoms must be followed carefully and a rapid diagnostic technique to inspect the VA system must be developed.1

Lessons
In our rare patient with a sudden-onset occipital headache, brain imaging studies returned no abnormal findings. Nonetheless, he died at home the next day. Precordial brain CT showed diffuse SAH. Pathologically, his death was due to the rupture of a blister-like VA aneurysm without dissection. We alert diagnosticians to the potential existence of such rare cases.

References

Disclosures
Dr. Kim reported personal fees from Daichi Sankyo and Nippon Zoki outside the submitted work.

Author Contributions
Conception and design: Kawauchi. Acquisition of data: Kawauchi, Inokuchi. Analysis and interpretation of data: Kawauchi. Drafting the article: Kawauchi, Kim. Critically revising the article: Kawauchi, Kim. Reviewed submitted version of manuscript: Kawauchi, Kim. Approved the final version of the manuscript on behalf of all authors: Kawauchi. Statistical analysis: Kawauchi. Administrative/technical/material support: Kawauchi. Kim. Study supervision: Kawauchi. Kim.

Correspondence
Masaaki Kawauchi: Chiba Shintoshi Rurban Clinic, Inzai, Chiba, Japan. aulora_king@chibashintoshi.or.jp.