Bath-Related Thunderclap Headache Associated with Subarachnoid and Intracerebral Hemorrhage

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Abstract-

Purpose: Bath-related thunderclap headache (BRTH) is a rare and usually benign condition. We report a case of episodic explosive thunderclap headache (TH) provoked by showering water, with the complications of cortical subarachnoid hemorrhages (SAH) and delayed intracerebral hemorrhage (ICH).

Case Report: A 56-year-old premenopausal woman, without chronic illness or headache history, suffered from 4 episodes of severe explosive TH within 11 days. Two of these attacks were provoked by hot water and 1 by cold water. A small acute SAH was found in the left high frontal cortex on brain computed tomography (CT) performed 7 days after the first attack (day 7). Brain magnetic resonance imaging (MRI) and angiography (MRA) on day 9 disclosed a new acute SAH in the right frontal cortex but with no apparent vasoconstriction. CT angiography (CTA) on day 12 first revealed vasoconstriction in the M2 segment of right middle cerebral artery (MCA), and found a new ICH in the right anterior frontal lobe. Conventional angiography on day 14 revealed partial remission of vasoconstriction with only mild short segmental narrowing at the proximal M1 segment of right MCA. The patient had no clinical neurological deficit. She was free of headache at day 11 when she started taking nimodipine.

Conclusion: Reversible cerebral vasoconstriction syndrome (RCVS) presented with BRTH is rare and is not always that benign as was once thought. The delayed ICH and the short-period of vasoconstriction in this patient extended our knowledge that the time course of the complications and the duration of vasospasm in RCVS could vary widely among patients. Nimodipine is probably effective in both relieving symptoms and reversing vasoconstriction.

Key Words: bath-related headache, thunderclap headache, reversible cerebral vasoconstriction syndrome

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INTRODUCTION

Headache induced by bathing is rare. The bath-related thunderclap headache (BRTH) syndrome was first introduced as a "benign hot bath-related headache" by Negoro et al., who reported 3 middle-aged Japanese women with paroxysmal thunderclap headache (TH) provoked on pouring hot water on themselves or soaking in a hot bath[(1)]. The headache was considered benign because it subsided spontaneously within 3 months,

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with no identifiable structural lesions found in the brain[1]. However, subsequent literature has stated that bath-related headache is not always a benign condition[2-4]. Well known complications include reversible posterior leukoencephalopathy and cerebral infarction, both of which are associated with transient multi-segmental intracranial vasoconstriction[2,4]. Wang et al. found that 60% of patients suffering from BRTH had cerebral vasospasms, fulfilling the diagnostic criteria of reversible cerebral vasoconstriction syndrome (RCVS)[4]. Here we reported a case of episodic intense TH provoked on bathing that was associated with transient cerebral vasoconstriction and complicated by multiple cortical subarachnoid hemorrhages (SAH) and a delayed intracerebral hemorrhage (ICH).

CASE REPORT

A 56-year-old Taiwanese woman with no known past medical history, presented with several bouts of severe abrupt headaches. She denied any history of prior significant headaches or any relevant family history. She was premenopausal, with irregular menses and did not receive hormone replacement therapy. The first headache occurred when she poured hot water over her head to wash out shampoo (day 1). The headache was sudden from the posterior neck to the occiput, reached its peak intensity within 1 minute, and was accompanied by mild nausea. No vomiting, phonophobia, photophobia, weakness or sensory symptoms were noted. The headaches lasted for approximately 2 hours and stopped after she received an intravenous analgesic at a local clinic, where her systolic blood pressure (SBP) was found to be 180 mmHg. She denied taking any medication within a month prior to this attack. The following day she took a shower, but not washing her hair, and no headache developed. The second headache occurred 2 days after the first episode (day 3), when a severe TH developed immediately after pouring hot water on her head. She self-recorded her SBP (190 mmHg) at home. The pain continued at an intense level for approximately 20 minutes, followed by a milder headache that lasted almost half a day. She refused hospitalization in our outpatient department, and therefore, 25 mg diclofenac twice daily was prescribed. Brain magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) was arranged.

Against medical advice, she continued to wash her hair in the following 3 days, without developing any headaches. Four days after the second episode, another headache occurred while she was talking to her boss (day 7). She felt stressful but denied anger, coughing, or walking into cold wind at that time. The headache was milder and developed more slowly than previous attacks (it took about 3 minutes to reach its peak intensity), and lasted for approximately half a day; she also recorded a SBP of 180 mmHg. When she presented to our emergency department, brain computed tomography (CT) showed a small, acute SAH in the left high frontal cortical sulcus (Figure 1A). She was discharged from the emergency department after the headache improved. Brain MRI and MRA was performed on day 9, and showed a subtle acute cortical SAH in the right frontal cortical sulcus (Figure 1B) but with no apparent cerebral ischemia or vasoconstriction (Figure 2A). The patient continued to shower but did not wash her hair. The fourth attack occurred 4 days after the third episode while she was just preparing to take a bath (day 11). A sudden explosive headache occurred when her hand touched cold water; her SBP was recorded at approximately 190 mmHg on that occasion. The severe intensity lasted for about 10 minutes with a milder headache remaining for hours. Due to the frequent thunderclap headaches, she finally agreed to be hospitalized in the neurology department on day 11.

At admission, she was afebrile and normotensive (SBP < 140 mmHg and diastolic blood pressure < 90 mmHg). Detailed neurological examinations were normal. Neither neck stiffness nor papilledema was recorded. Treatment was started at admission, with oral nimodipine, 60 mg every 4 h. Findings from laboratory studies, including antinuclear antibody, anticardiolipin antibody, C3, C4, antithrombin, protein C, protein S, and homocysteine, were within normal limits. CTA on day 12 (the second day of admission) revealed a segmental narrowing of the M2 segment of right middle cerebral

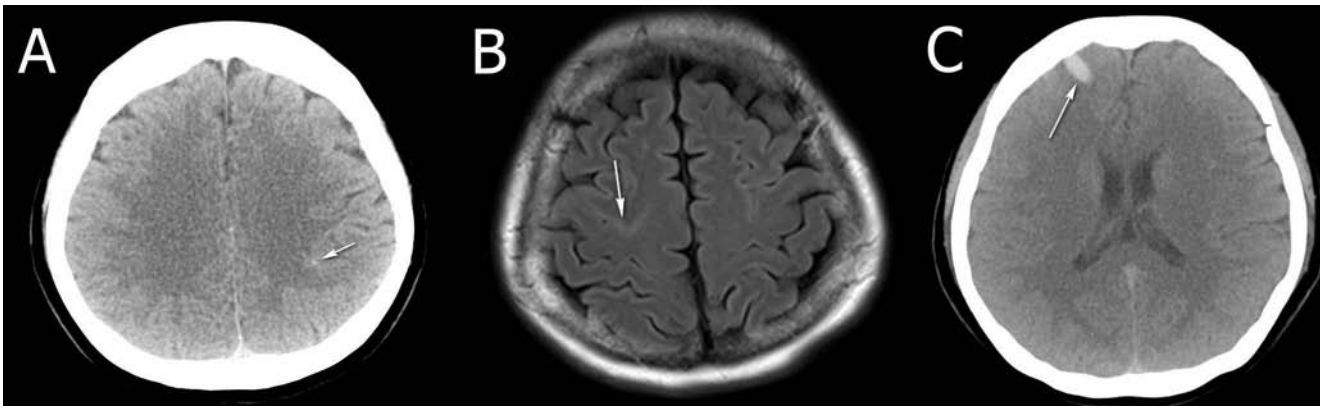
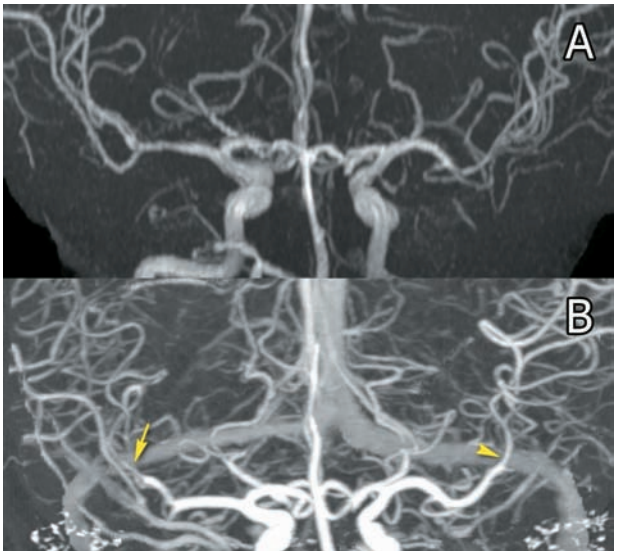


Figure 1. Hemorrhagic complications on serial neuroimaging. (A) CT on day 7: a small, acute SAH in the left high frontal cortical sulcus (B) MRI on day 9: Axial T2-weighted fluid attenuated inversion recovery image showed a subtle acute cortical SAH in the right frontal cortical sulcus (C) CTA on day 12: a new 11 ×9 ×15 mm-sized ICH in the right anterior frontal lobe.



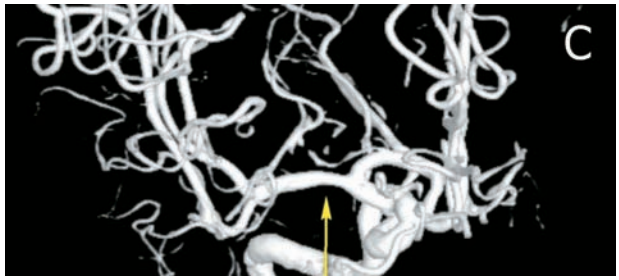


Figure 2. Serial cerebral vascular imaging. (A) MRA on day 9: An intracranial 3D time-of-flight MRA showed no apparent vasoconstriction. (B) Dual energy CTA on day 12: A 3D intracranial CTA with maximum intensity projection technique showed a segmental narrowing of the M2 segment of right middle cerebral artery (MCA, arrow) while left MCA was normal (arrowhead). (C) Digital subtraction angiography on day 14: The 3D rotational angiography of volume rendering modes showed resolution of the right cerebral arterial vasoconstrictions, but with a residual mild short segmental narrowing of the proximal M1 segment of the right MCA.

artery (MCA) (Figure 2B) and found a new 11 ×9 ×15 mm-sized ICH in the right anterior frontal lobe (Figure 1C), which was not seen in the previous MRI performed on day 9. ICH did not result in corresponding symptoms or cause seizures. Transcranial color-coded sonography on day 12 revealed normal flow velocities and pulsitivity indices bilaterally in the anterior cerebral artery (ACA), MCA (both in M1 and M2 segments), posterior cerebral artery (PCA), and the distal internal carotid artery (ICA). Conventional carotid angiography on day 14 showed resolution of the right cerebral arterial vasoconstrictions, but with a residual mild short segmental narrowing of the proximal M1segment of the right MCA (Figure 2C). No aneurysms, vascular malformations, or tumors were found. She experienced no further headaches during hospitalization. The temporal relationship between clinical symptoms and imaging findings was illustrated in Figure 3.

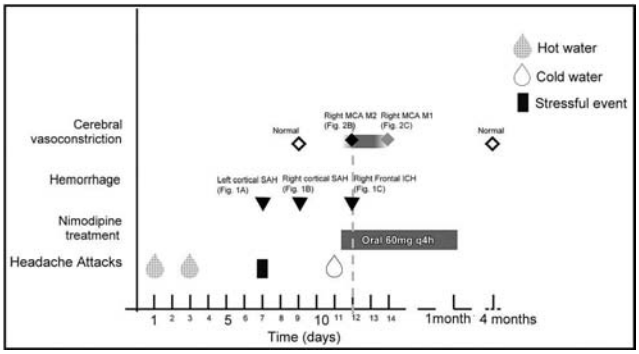


Figure 3. Illustration of the temporal relationship between clinical symptoms and imaging findings.

The dose of nimodipine was tapered and completely discontinued 4 weeks after the initial prescription. She remained headache free in the following months. A follow-up MRA, 4 months after the first presentation showed complete remission of the cerebral vasoconstrictions.

DISCUSSION

Our patient, a middle-aged woman with no history of prior headache, experienced 4 attacks of explosive TH within 11 days and had the complications of SAH and delayed ICH.

Three of the four attacks were associated with water contact: two were provoked by pouring hot water over her head and one by touching cold water. The duration of these severe headaches was between 10 min and 2 h, with or without a milder headache remaining. The features and clinical courses were both compatible with previously reported BRTH[1,3-6]. All published cases to date have been women, most have been middle-aged and menopausal[4], and the duration has ranged from 6 days to 3 months[3,4]. Both cold and hot water showering have been associated with triggering headaches[3,4]. In patients with BRTH, non-bath related triggers have also been reported, including brushing teeth with cold water, walking into cold wind, exertion, straining at stools, coughing, anger, sexual intercourse, and singing[4,7]. The third attack of headaches in this patient could not be classified as a TH due to its longer onset (>1 min), although the patient described the pain as of similar characteristics to the other attacks. We speculate that this attack was triggered by the stressful situation of talking to her boss.

Patients with BRTH can be diagnosed as having RCVS if vasoconstriction is seen, or be diagnosed as having primary TH if there is no any identifiable pathology[4]. Our patient fulfilled the diagnostic criteria of RCVS apart from the lack of cerebrospinal fluid (CSF) analysis[8]. However, the requirement for CSF analyses in the diagnosis of RCVS has been challenged in cases where there are both multiple thunderclap headaches and typical angiographic features[9]. In spite of the lack of a CSF report, meningitis was unlikely since there was no fever or neck stiffness. Other differential diagnoses were excluded via neuroimaging, including: aneurysm, dissection of intra- or extra-cranial vessels, venous sinus thrombosis, pituitary apoplexy, spontaneous intracranial hypotension. Primary angiitis of the central nervous system is highly unlikely because: the classic features of episodic thunderclap headaches were suggestive of RCVS; headaches were not progressive and had a monophasic course; and the fact that there was a complete resolution of vasoconstriction without the need to administer immunosuppressants[9-11].

Hemorrhagic complications in RCVS had been widely reported, including SAH, focal intracranial hemorrhage, and very rarely, subdural hemorrhage[10,12]. However, diagnosis of vasoconstriction occurred either concomitant with, or after the diagnosis of hemorrhage[12-14], making it difficult to prove a causal relationship between SAH and vasoconstriction[15]. In this report, vasoconstrictions and hemorrhages in the right frontal lobe that were in the supplying territories of the contracted arteries, were diagnosed in the same image session on day 12. Whilst the cortical SAHs were found early, the ICH must have developed between day 9 and 12 after symptom onset. Based on the temporal and spatial relationships of the right frontal ICH and vasoconstrictions, it is reasonable to conclude that intracranial bleeding is probably a result of angiopathy, but not a trigger[12].

Another striking feature of the present patient was the delayed emergence of ICH on day 12. The mean

delay from headache onset to the diagnosis of ICH and cortical SAH were 2.2±2.5 days and 4.6±4.3 days, respectively[12]. This is much longer than has previously been recognized. Our case emphasized the possible delayed occurrence of ICH in the second week after headache onset.

Serial cerebral angiographies of this patient uncovered a trend of distal to proximal vasoconstriction in the right MCA (Figure 2B, 2C). Previous literature has reported similar findings in RCVS patients with newer vasoconstrictions often affecting more proximal vessels[16,17]. Although caliber irregularities are often bilateral and diffuse, the typical findings of a string of beads may only be seen in one or more cerebral artery[8,10]. The demonstration of vasoconstriction in this patient was most clearly visualized in the dual energy CTA, the advantage of which in the diagnosis of RCVS, has been reported previously[18].

Another unusual finding is that the patient had a relatively rapid remission of vasoconstriction as compared with previous BRTH reports[2]. Vasoconstriction had almost completely resolved by the third day of oral nimodipine administration (day 14). Studies have reported that the mean duration of partial resolution in RCVS, using MRA as the diagnostic tool, was 33±24 days, with 11 days being the shortest time[19]. We do not know whether the rapid resolution of vasoconstriction was a variation of the natural course or if it was due to treatment with nimodipine.

The blood pressure of our patient became elevated to approximately 180 mmHg each time she suffered the intense headaches. She had no history of hypertension and her blood pressure during hospitalization was normal. An elevated blood pressure has been reported in more than one third of patients with RCVS[12,17,19,20], and is associated with a higher risk of posterior reversible encephalopathy syndromes (PRES)[20], but not an increased risk of hemorrhagic complications[12]. This case had no clinical or radiological evidence of PRES.

Nimodipine was prescribed on day 11, and the patient became headache free. The effective rates of nimodipine in terminating headache has been reported at 84% in patients with BRTH[4] and 59%-83% in patients with RCVS[17,19]. There have been no randomized placebo-controlled trails to support this widely employed treatment. When comparing the clinical outcomes using a modified Rankin scale, nimodipine offers no benefit except for the relief of symptoms[21]. Nimodipine probably does not affect the time course of cerebral vasoconstriction[10]. Further comprehensive studies are needed to determine the effectiveness of nimodipine, in terms of symptom relief, regression time of vasoconstriction, and the rate of various complications associated with RCVS.

The pathophysiology of BRTH is poorly understood. Possible etiological explanations include autonomic neurovascular reflexes triggered by temperature changes[7]; increased sympathetic tone during standing while showering[4]; and estrogen deficiency or fluctuation at the menopause or post-medical oophorectomy[4,6]. Also, ethnic factors may play a role, as there is a higher incidence in Eastern populations than in the West[3-5].

BRTH is rare and is not always a benign condition as was once thought. The delayed ICH and the short-period of vasoconstriction in this patient extended our knowledge that the time course of the complications and the duration of vasospasm in RCVS could vary widely among patients. Nimodipine is probably effective in both relieving symptoms and reversing vasoconstriction.

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