Short report

Reversible cerebral vasoconstriction syndrome after aneurysmal clipping

Masaya NAKAGAWA, Satoru TAKEUCHI, Sho SATO, Yuhei SUZUKI,
Daichi HAGITA, Yohei Otsuka, Satoshi TOMURA, Terushige TOYOOKA,
Arata TOMIYAMA and Kojiro WADA

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Abstract: Reversible cerebral vasoconstriction syndrome (RCVS) is characterized by thunderclap headache and reversible cerebral vasoconstriction. We report a rare case of RCVS after aneurysmal clipping surgery. The patient was a 50-year-old woman with a history of hypertension who had developed a sudden-onset severe headache one week previously. Brain fluid-attenuated inversion recovery (FLAIR) magnetic resonance imaging (MRI) revealed cortical SAH. Three-dimensional computed tomography angiography (3D-CTA) demonstrated small aneurysms in the anterior communicating artery and the left internal carotid artery (ICA). Aneurysmal clipping was successfully performed. The intraoperative findings revealed that both aneurysms were unruptured. Postoperative brain 3D-CTA showed no residual aneurysms without occluded vessels. However, the patient presented with left-sided hemiparesis and sensory disturbance on day 2. Brain diffusion-weighted MRI demonstrated hyperintense lesions in the right medial parietal lobe. Brain 3D-CTA revealed multiple narrowing of the cerebral arteries. Based on the clinical course and the presence of vasoconstriction, RCVS was suspected, and continuous calcium channel blocker treatment was administered for two weeks. Follow-up 3D-CTA showed the disappearance of segmental narrowing. She was therefore diagnosed with definite RCVS. Her symptoms gradually improved, and she was discharged with mild left-sided sensory disturbance one month later.

Key words: reversible cerebral vasoconstriction syndrome ／ stroke ／ subarachnoid hemorrhage ／ cerebral infarction

Introduction

Reversible cerebral vasoconstriction syndrome (RCVS) is characterized by thunderclap headache and reversible cerebral vasoconstriction. The cause is unknown. It is reported that the frequency of RCVS in women is twice that in men. Complications may include stroke (e.g., subarachnoid hemorrhage [SAH], cerebral hemorrhage, or cerebral infarction). We herein report a rare case of RCVS after aneurysmal clipping.

Case

The patient was a 50-year-old woman with a history of hypertension who developed a sudden-onset severe headache one week previously, and was admitted to our institution because it had not stopped. The patient had no prior history of migraine or other thunderclap headache, and no history of vasoconstrictive drug use. On admission, she was alert. There were no focal neurological signs. Her vital signs were as follows: temperature, 36.4℃; blood pressure, 158/92 mmHg; pulse, 93 bpm; respiratory rate, 22 bpm. The endocrinology test for detecting

Department of Neurosurgery, National Defense Medical College, Tokorozawa, Saitama 359-8513, Japan.

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secondary hypertension was not performed. Brain fluid-attenuated inversion recovery (FLAIR) magnetic resonance imaging (MRI) showed cortical SAH (Fig. 1a). 3D-CTA demonstrated small aneurysms in the anterior communicating artery and the ophthalmic segment of the left internal carotid artery (ICA) (Figs. 1b, c).

Aneurysmal clipping was successfully performed. The intraoperative findings revealed that both aneurysms were unruptured. Postoperative brain 3D-CTA showed no residual aneurysms without occluded vessels (Fig. 1d). However, the patient presented with left-sided hemiparesis and sensory disturbance on day 2.

Brain diffusion-weighted MRI demonstrated hyperintense lesions in the right medial parietal lobe (Fig. 2a).

Brain 3D-CTA revealed multiple narrowing of the cerebral arteries, especially the right anterior cerebral artery (ACA), which was occluded in the A2 segment (Fig. 2b). Based on the clinical course and the presence of vasoconstriction, she was suspected to have RCVS, and received continuous calcium channel blocker treatment for two weeks. Follow-up 3D-CTA on day 15 showed the disappearance of segmental narrowing, and recanalization of the right ACA (Fig. 2c). She was therefore diagnosed with definite RCVS. Her symptoms gradually improved, and she was discharged with mild left-sided sensory disturbance one month later.

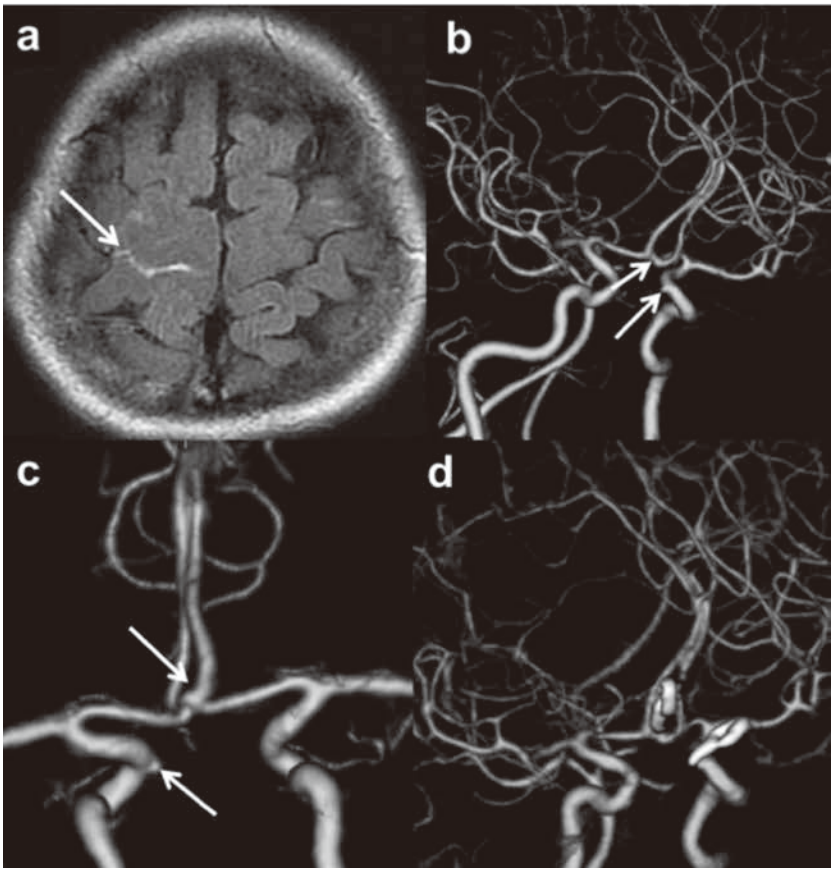

Figure 1.
a: A brain fluid-attenuated inversion recovery magnetic resonance image obtained on admission showed cortical subarachnoid hemorrhage (arrow).
b, c: Brain three-dimensional computed tomography angiography (b, right oblique view; c, postero-anterior view) performed on admission demonstrated small aneurysms in the anterior communicating artery and the ophthalmic segment of the left internal carotid artery (arrows).
d: Postoperative brain three-dimensional computed tomography angiography (right oblique view) showed no residual aneurysms. Vessel occlusion was not observed.

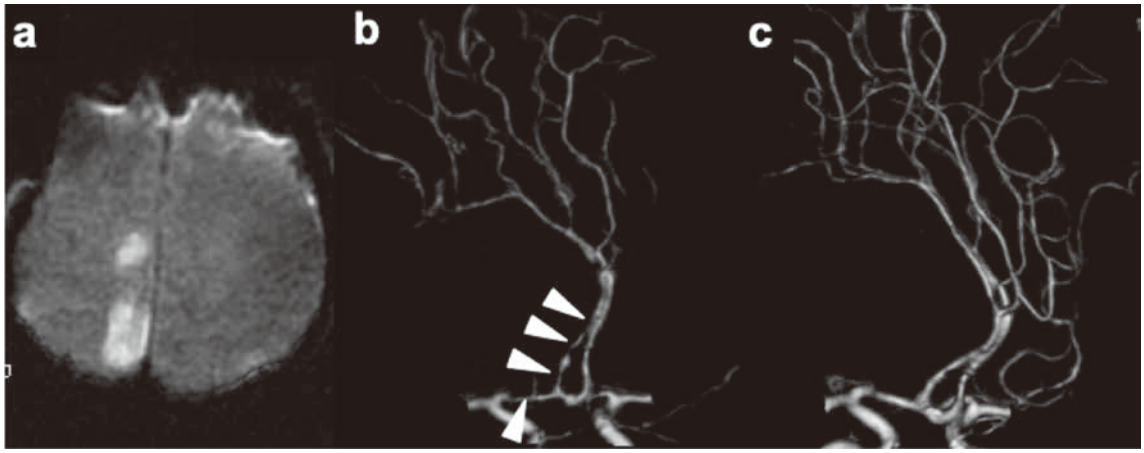

Figure 2.
a: A brain diffusion-weighted magnetic resonance image obtained on day 2 demonstrating hyperintense lesions in the right medial parietal lobe.
b: Brain three-dimensional computed tomography angiography (right oblique view) revealed multiple narrowing of the cerebral arteries, especially the right anterior cerebral artery, which was occluded in the A2 segment (arrowheads).
c: Follow-up three-dimensional computed tomography angiography (right oblique view) on day 15 showed the disappearance of segmental narrowing and recanalization of the right anterior cerebral artery.

Discussion

RVCS is characterized by sudden-onset headache, with or without neurological deficits associated with multifocal, reversible narrowing of the cerebral arteries on angiography [1-3]. RCVS is often associated with several complications. Among these, non-aneurysmal cortical SAH, intracerebral hemorrhage and reversible posterior leukoencephalopathy syndrome mainly occur within the first week, and transient ischemic attacks and cerebral infarction occur later [1-3]. RCVS is traditionally considered to have a monophasic and benign clinical course [1]. In contrast, Katz et al. recently advocated that clinical worsening after a diagnosis of RCVS is more common than has been previously believed [3]. In addition, in a series of 67 patients reported by Ducros et al., at least one patient (1.6%) who presented with a hemorrhagic event experienced a subsequent delayed ischemic event [2]. Idiopathic RCVS is most common [4]. Other associations with RCVS have been reported that medications especially vasoactive drugs and immunosuppressants, pregnancy, migraine, blood products, and catecholamine-secreting tumors. After neurosurgical procedures, RCVS can also occur [5]. It has been reported that the majority of non-traumatic SAH under 60-year-old are caused by RCVS [6]. In our case, the intraoperative findings revealed that both aneurysms were unruptured, and therefore we considered that RCVS might have caused SAH despite no findings of vasoconstriction on admission. Careful attention should be paid to the possibility that RCVS can cause both hemorrhagic events and subsequent ischemic event. The treatment of RCVS is controversial [1-3]. Calcium channel blockers have been the most common treatment for affected patients, and a calcium channel blocker was effective in our case.

Conclusion

Careful attention should be paid to the possibility that RCVS may occur after neurosurgical procedures.

Conflicts of Interest

The authors have no conflicts of interest directly relevant to the content of this article.

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開頭クリッピング術後に発症した可逆性脳血管攣縮症候群の１例

中川政弥，竹内　誠，佐藤　翔，鈴木悠平，萩田大地，大塚陽平，戸村　哲，
豊岡輝繁，富山新太，和田孝次郎

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要旨：可逆性脳血管攣縮症候群（以下，RCVS）は雷鳴様頭痛と可逆性の脳血管狭窄を特徴とし，時に出血性あるいは虚血性脳合併症を併発することがある。今回我々は，開頭クリッピング術後に，RCVSによる脳梗塞を認めた症例を経験した為，文献的考察を加えて報告した。症例は，高血圧の既往のある50歳女性。１週間前から継続する頭痛を主訴に入院した。頭部MRIにて右頭頂葉に薄いくも膜下出血を呈しており，脳 3D-CTAで前交通動脈と左内頸動脈に小脳動脈瘤を認めた。開頭クリッピング術を施行したが，術中所見は未破裂瘤であった。手術は問題なく終了し，術後の脳 3D-CTAでは残存瘤や脳主幹動脈の閉塞を認めなかったが，術後２日目に左片麻痺が出現した。頭部MRIで右頭頂葉内側に急性期脳梗塞があり，脳 3D-CTAでは，右前大脳動脈を中心に，脳血管攣縮を多発性に認めた。経過及び画像所見からRCVSを疑い，カルシウムチャネル遮断薬の投与を２週間継続し，脳 3D-CTAにて脳血管攣縮の改善が確認出来た。RCVSは単一の症状を呈して，良性の臨床経過を辿ることが多い。RCVSの発症リスクとして，血管作動薬，免疫抑制薬，分娩，片頭痛等の報告がある。開頭手術もRCVSの一因とされており，開頭術後管理において留意する必要がある。

索引用語：　可逆性脳血管攣縮症候群　／　脳卒中　／　くも膜下出血　／　脳梗塞

防衛医科大学校脳神経外科学講座