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Full Length Research Paper

Symptomatic intracranial capillary telangiectasia in a patient with contralateral common carotid artery dissection

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Intracranial capillary telangiectasia (ICT) is a rare benign disease with few clinical reports, as most capillary telangiectasias locate in the brainstem, small, asymptomatic and angiographically invisibale. We present a case of intracranial capillary telangiectasia with seizure and hemiplegia. Cranial magnetic resonance imaging (MRI) studies revealed right frontal ischemic lesion. Digital subtraction angiography (DSA) demonstrated right hemispheric capillary telangiectasia of ipsilateral frontal ischemic lesion and left common carotid artery dissection.

Key words: Capillary telangiectasia, acute ischemic stroke, digital subtraction angiography.

INTRODUCTION

Intracranial capillary telangiectasia (ICT) is a benign asymptomatic, angiographically invisible disease with few clinical reports (Sayama et al., 2010). It is difficult to diagnosis clinically. This report presents a case of ICT presenting with seizure and hemiplegia without neck trauma before or a history of excessive force activity. Digital subtraction angiography (DSA) demonstrated the right hemispheric CT and contralateral common carotid artery dissection.

CASE REPORT

A 17-year-old Chinese boy was admitted to our hospital with (due to) seizure. For six days he had been suffering from seizure after drinking and was found to have a weakness in the left upper and lower limbs after the second seizure attack. The patient was non-smoker and past medical history was unremarkable.

General physical examination showed no abnormalities, blood pressure was 150/90 mmHg in both arms. Neurological examination revealed left weakness in the left arm and leg with diffusely increased deep tendon reflexes. The National Institute of Health Stroke Scale (NIHSS) score was 7. There was no major artery bruit.

Routine blood laboratory, immunology, coagulation and cerebral spinal fluid (CSF) laboratory analysis were normal. Electrocardiography and chest X-ray were unremarkable. Electroencephalography did not reveal any other epileptogenic focus in the brain. Cranial MRI revealed a right frontoparietal large infarction and small left frontal large infarction (Figure 1). Transthoracic echocardiography showed normal findings without evidence of possible cardiac sources of emboli. Transcranial eco-colour doppler revealed absence of flow in the left cerebral area due to extracranial internal carotid artery (ICA)-related lesion. Collateral supply through the anterior communicating part of the posterior cerebral artery and the postcommunicating part of the posterior cerebral artery to the middle cerebral artery (MCA) was

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Abbreviations: ICT, Intracranial capillary telangiectasia; **MRI,** magnetic resonance imaging; **DSA,** digital subtraction angiography; **NIHSS,** National Institute of Health Stroke Scale; **CSF,** cerebral spinal fluid; **SWI,** susceptibility weighted image; **MCA,** middle cerebral artery; **ICA,** internal carotid artery.

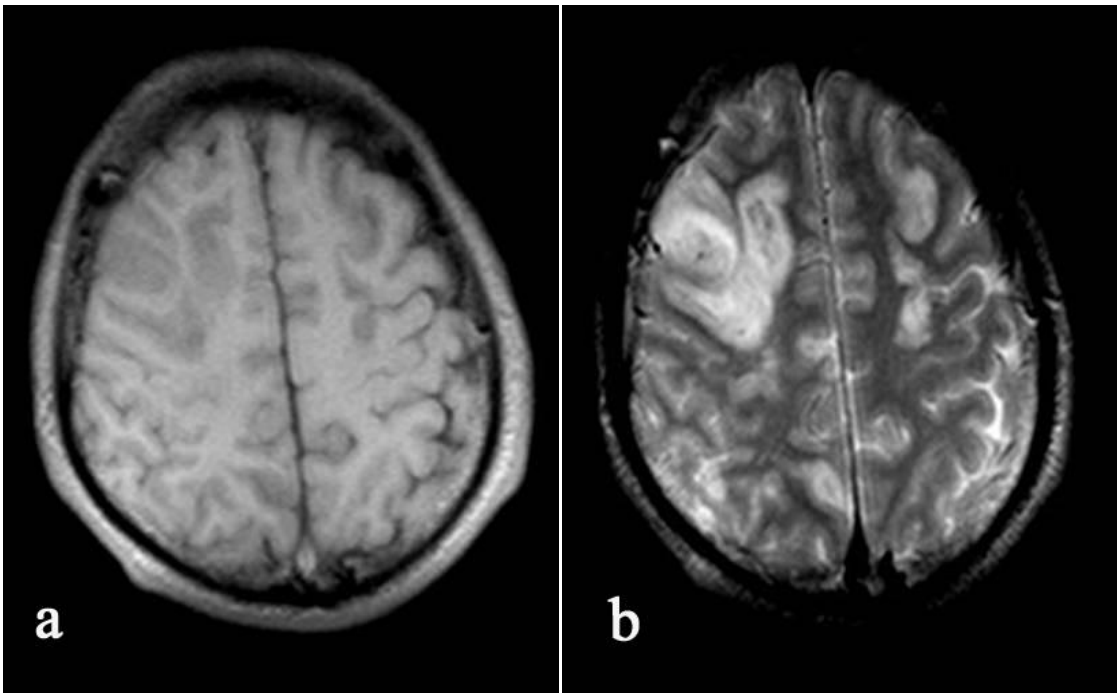

Figure 1. Axial cranial MRI. T1, T2 weighted images indicate abnormal long-T1, long-T2 signals in the right frontal lobe. a, T1-weighted images, b; T2-weighted images.

demonstrated. Carotid ultrasonography showed atherosclerotic plaque of the right internal carotid tree on both sides. There was a left spontaneous common carotid dissection with a thrombosis, resulting in the occlusion in the origin of the left internal carotid artery (Figure 2). Brain DSA demonstrated an occlusion of left common carotid artery (Figure 3a). DSA showed abnormal capillary mission of the right frontal lobe at capillarious phase, abnormal corpus knee artery imaging at capillarious and venous phase (Figure 3b, c and d).

Because there are no randomized trials to be compare with the therapy efficacy of anticoagulant and antiplatelet, and non-randomized test analysis did not find any difference between the two treatments. In another hand, the patient lived in a relatively remote place where is not convenient to test the INR and his familiy refused to use warfarin, so the patient was put on single-drug

So the patient was put on single-drug anti-epileptic and aspirin treatment, and accepted rehabilitation. He was discharged 12 days after admission, NIHSS score was 3. He has no recurrent ischemic events and seizure attacks during one year's follow-up.

DISCUSSION

Cerebral vascular malformations are classified into arteriovenous malformation, developmental venous anomalias, cavernomas and capillary telangiectasias (CT). CT account for 16-20% of all intracerebral vascular malformations (Sayama et al., 2010). Most CT commonly locate in the brainstem, especially the pons (Scaglione et al., 2001), but may also be found in the cerebral lobes (Castillo et al., 2001). Most of the lesions are incidental findings, as these malformations are generally small, clinically asymptomatic, angiographically invisible and pathological confirmation is absent. Capillary telangiectasias may occasionally be symptomatic and increasingly recognized by contrast-enhanced MRI imaging (Lee et al., 1997).

Here we report a case of cerebral CT recognized by cranial DSA. Our patient suffered from an acute stroke which presented with seizure and hemiparesia after alcohol drinking. Except for an ischemic lesion in the frontal-temple lobe, standard MRI did not show any typical characteristic of capillary telangiectasias. Unfortunately, our patient did not get a susceptibility weighted image (SWI) which can increase diagnostic accuracy as reported on literatures (Yoshida et al., 2006; Finkenzeller et al., 2010). But DSA revealed abnormal capillary at capillarious phase of the right ICA, however. These findings suggest that this abnormal lesion is a CT and there is no report about simultaneous of telangiectasia or carotid dissection before.

In our patient, the occlusion of the left ICA with good collateral circulation was probably not the key cause of the acute infarction of right frontal-temple lobe. The occlusion of the left ICA just resulted in a small infarction

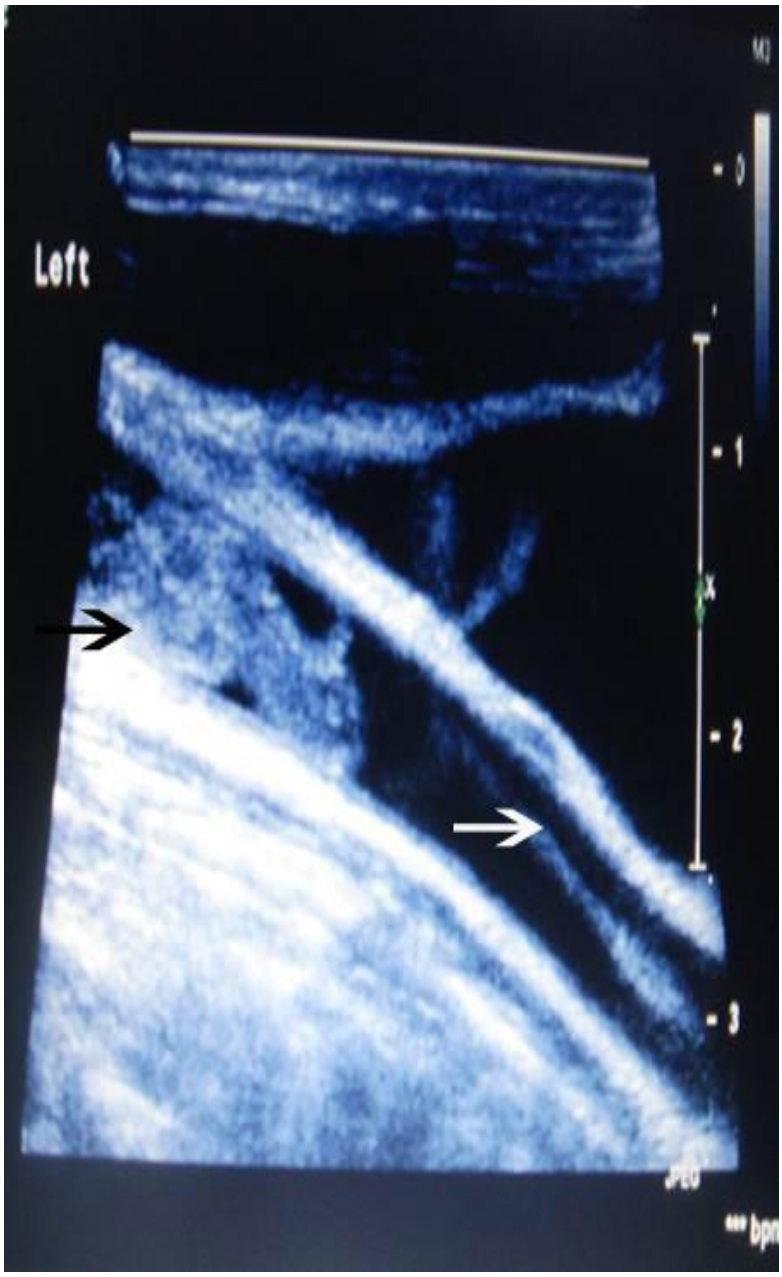

Figure 2. Carotid ultrasonography indicates a left spontaneous common carotid dissection (white arrow) with a thrombosis (black arrow), resulting in the occlusion in the origin of the left internal carotid artery.

of the left frontal lobe without corresponding symptoms. Moreover, we observed abnormal capillary at capillarious phase of the right ICA on DSA. CT was causal associated with the ipsilateral superficial ischemic lesion due to abnormal hemodynamic. The right frontal-temple ischemia is responsible for the symptoms of the patient.

Taken together, our case demonstrates seizure and hemiplesia corresponding to the ischemic lesion due to frontal capillary telangiectasia. Although no pathologic confirmation is available in our patient, the vascular malformations were recognized by DSA. DSA could provide supportive diagnosis information of capillary telangiectasia. ICT is a rare disease about small cerebral vascular malformations which may form congenitally, while the cause of carotid artery dissection may be spontaneous and traumatic. The patient denied any trauma and other acquired causes that lead to dissection.

The existence of telangiectasia and carotid dissection in

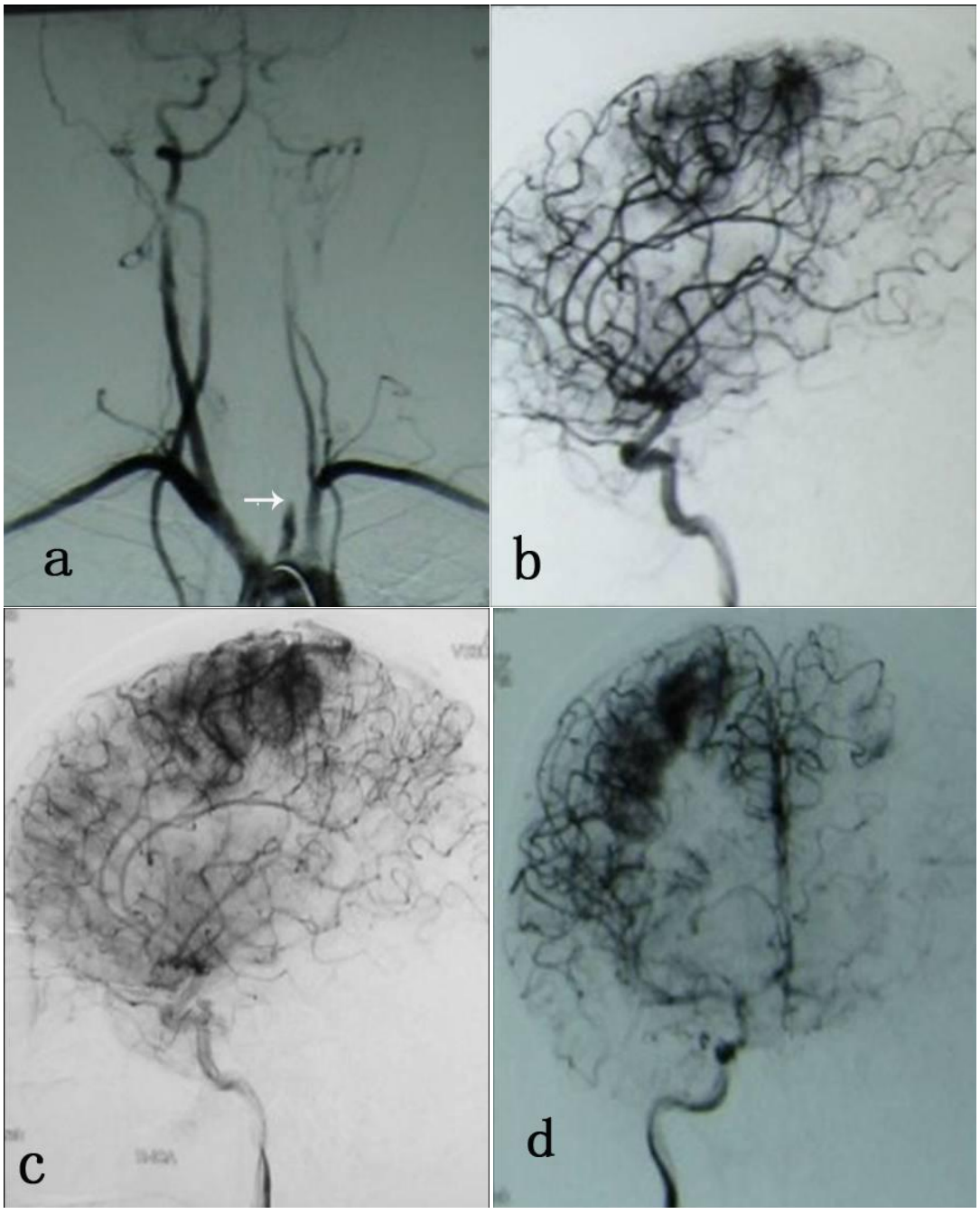

Figure 3. Cranial DSA. a, anteroposterior DSA shows occlusion in the left commom carotid artery(white arrow). b, c and d) DSA demonstrates the right fronto-parietal diffusion abnormal capillary mission in early and late phases. Note abnormal corpus knee artery imaging at capillarious and venous phase without venous drainage. (b: early phases, c: late phases, d: anteroposterior view).

the patient suggested that perhaps they are developed by congenital causes.

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