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Case Report

Hemorrhagic presentation without venous infarction caused by spontaneous thrombosis of developmental venous anomaly and angiographic change after treatment

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ABSTRACT

Developmental venous anomalies (DVAs) have been known benign anatomical variations and generally do not cause symptoms. Although several cases of venous infarction caused by DVA thrombosis have been reported, it not common in itself. But only hemorrhagic presentation in DVA is extremely rare. We describe a case with large intracerebral hemorrhage (ICH) without venous infarction caused by DVA thrombosis and angiographic change of successful treatment.

1. Introduction

We have known about various vascular anomalies of the central nerve system including arteriovenous malformations (AVMs) and Developmental venous anomalies (DVAs) are a fairly commonly reported entity. However, they have been known as mostly benign anatomical variations that generally cause no symptoms [1]. DVAs occur during the developmental process of the venous system, by retention of the primitive embryological medullary vein [2]. It is well known that it represents the shape of the caput medusa, composed by one longer drainage vein. Although venous infarction caused by DVA is uncommon, several cases have been reported and most cases have been known to be associated with venous thrombosis of the draining vein [3]. But presentation with only hemorrhage in DVA is extremely rare. We are reporting a case of large intracerebral hemorrhage (ICH) without venous infarction caused by DVA thrombosis and would like to discuss use of anticoagulant in treatment of symptomatic DVA thrombosis with ICH.

2. Case report

A 27-year-old woman without past any remarkable medical history or medication was admitted with a general tonic clonic seizure and sudden onset decreased consciousness. Neurological examination showed drowsy consciousness with language and motor impairments. Computerized tomography (CT) scan revealed ICH in the left frontal lobe with intraventricular hemorrhage (IVH) (Fig. 1A). CT angiography showed venous dilatation in the anterior portion of ICH (Fig. 1B). Conventional and 3D rotational angiogram defined a large DVA collecting numerous small veins, contributing to the caput medusae shape in the venous phase. But the main draining vein of DVA did not drain into the superior sagittal sinus (SSS) and thrombosis was found in several disconnected areas regarding the SSS (Fig. 2A). Evaluation for hypercoagulable state was performed, the blood laboratory studies for thrombosis were either negative or within normal range: antiphospholipid, antinuclear and anticardiolipin antibodies, antithrombin III, lupus anticoagulant, factor V, protein C and S activity, and homocysteine. The patient had no history of oral contraceptive drugs. Immediate anticonvulsant therapy and intracranial pressure control were initiated. Due to the acute ICH, use of anticoagulants was of major concern. But with the assumption that without worsening of thrombosis, additional hemorrhage would not be a problem, systemic heparinization was started. This decision is based on the concept that anticoagulant therapy may be considered for venous sinus thrombosis with intracranial hemorrhage based on case series, although it has not been established anticoagulant is appropriate treatment for DVA thrombosis. Ten days later, follow-up cerebral angiography revealed the much decreased thrombosis and good drainage to the DVA (Fig. 2B). During use of anticoagulant, follow-up CT scan was performed at intervals of several days. Fortunately, the size of the hematoma was not increased and additional hemorrhage was not occurred (Fig. 3). The patient's neurological symptoms were gradually improved and she was discharged without any neurologic deficits. Anticoagulants and anticonvulsants were maintained for 6 months after discharge and neurologic changes or new symptoms were not observed after drug withdrawal. Initial brain magnetic resonance imaging (MRI) showed

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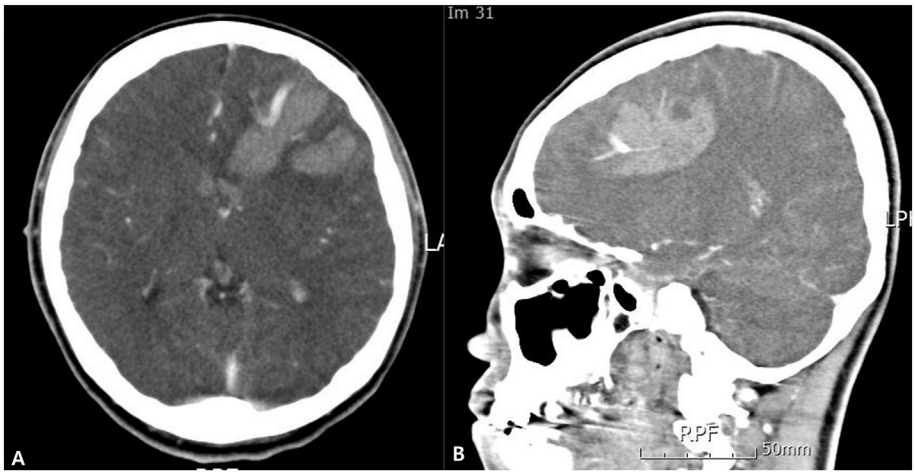

Fig. 1. A. Initial CT scan revealed ICH in the left frontal lobe and IVH.
B. CT angiography showed venous dilatation in anterior portion of ICH.

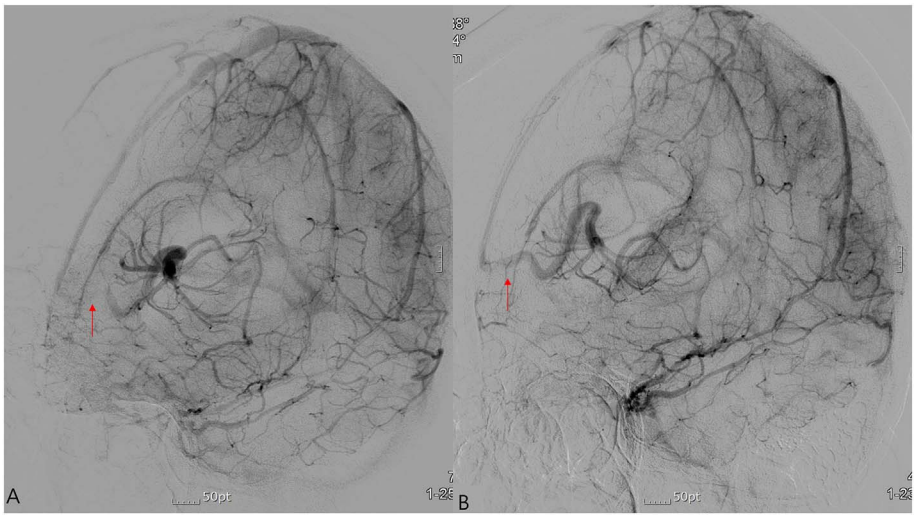

Fig. 2. A. Conventional and 3D rotational angiogram defined a large DVA collecting numerous small veins, contributing to the caput medusae shape in the venous phase. But the main draining vein of DVA did not drain into the superior sagittal sinus (SSS) and thrombosis was found in several disconnected areas regarding the SSS (red arrow).
B. Ten days later, follow-up cerebral angiography revealed that thrombosis in DVA was much decreased and DVA was connected to SSS and blood flow was well drained through DVA (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

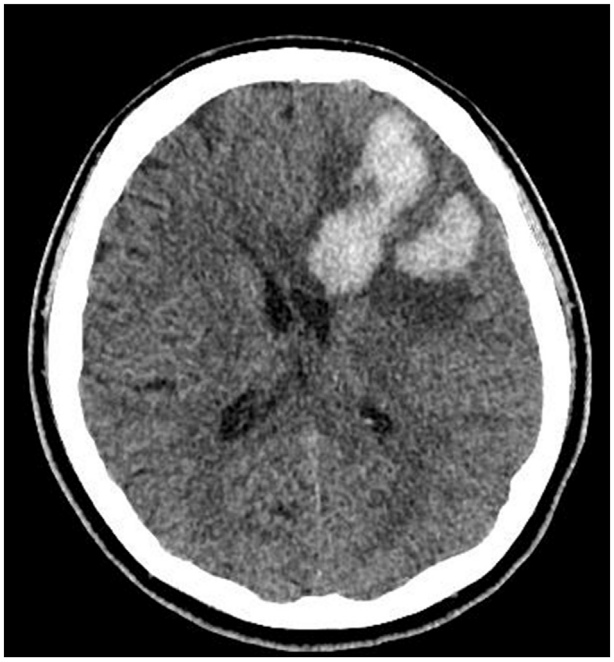

Fig. 3. Five days later after systemic heparinization, follow-up CT showed hematoma was not increased and additional hemorrhage was not occurred.

the same hemorrhagic findings as CT (Fig. 4A and B). There was no definite restriction, high signal in diffusion image (Fig. 4C). Although absence of restricted diffusion, venous infarction cannot be completely ruled out as a possible etiologic component of dominant hemorrhagic pathology. But we assumed that, if the hemorrhage had been secondary to transformation of the initial infarction, there would have been signs of infarctions in the original site, but in this patient, there was no such evidence. The opinion of the radiologist was the same as ours. In CT after 5 months, only small cerebromalatic change was visible in previous hematoma area and there was no evidence of cerebral infarction (Fig. 4D). If the hemorrhage were secondary to cerebral venous infarction, there would have been a broader cerebromalatic change involving cortical and subcortical area in follow up CT after 5 months. These findings suggest that it was a pure ICH without venous infarction rather than hemorrhagic transformation in infarcted area.

3. Discussion

DVAs represent a dilatation of a cerebral draining vein, which acts as a collector for small venules; DVAs are often found incidentally on examination and are generally considered as benign congenital vascular malformations, present in approximately 2.6% of the general population [4]. Generally, DVAs occur during the developmental process of the venous system, by retention of the primitive embryological medullary vein [5]. DVAs present a characteristic appearance on CT and MR studies, and seldom require cerebral angiography [6]. DVAs are rarely found in CT scans without contrast. After administration of