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Reversible cytotoxic edema associated with dural arterio venous fistula: a case report

Süleyman Men[a,*], Fethi İdiman[b], Aytaç Gülcü[a], Burcu Örmeci[b], Burak Paköz[b]

[a] *Department Radiology, Medical School, Dokuz Eylül University, İnciraltı İzmir, TR-35340 İzmir, Turkey*
[b] *Department of Neurology, Medical School, Dokuz Eylül University, İnciraltı İzmir, TR-35340 İzmir, Turkey*

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Abstract

Dural arterio venous fistulas with restricted antegrade venous flow and reflux into a dural sinus or cortical veins lead to intracranial venous hypertension. This type of venous hypertension in return may cause parenchymal damage with mechanisms similar to those proposed for venous infarction. A 67-year-old female presented with mixed aphasia and loss of consciousness that had progressed within the last 24 h. T2 and fluid attenuated inversion recovery weighted magnetic resonance images showed prominent cortical veins and diffuse white matter hyperintensity in the temporal, parietal and occipital lobes. The hyperintense areas also showed restricted diffusion on the diffusion weighted imaging. Angiography confirmed dural arteriovenous fistula of the torcular herofili, severe stenosis of the right transverse-sigmoid sinus junction, occlusion of the left sigmoid sinus and reversal of the flow in the superior sagittal sinus. The stenotic sinus was dilated with balloon angioplasty, while the fistula was treated with particle embolization. This report emphasizes on unusual coexistence of cytotoxic edema with dural arteriovenous fistula and its reversal after elimination of venous outflow obstruction.

Keywords: Dural arterio venous fistula; Diffusion weighted imaging; Cytotoxic edema

Both experimental and clinical studies have shown that diffusion weighted imaging (DWI) is very sensitive in the diagnosis of acute arterial stroke [1]. However, relatively little is known about the significance of DWI in the neurologic disorders resulting from the cerebral venous circulation. Venous sinus thrombosis and its consequences have been studied with DWI to some extent. It has been shown that both cytotoxic and vasogenic edema play a role in the pathogenesis of the cerebral damage secondary to venous thrombosis [2–4]. Dural arterio venous fistulas (DAVFs) with restricted antegrade venous flow and reflux into a dural sinus or cortical veins lead to intracranial venous hypertension. This type of venous hypertension in return may cause parenchymal damage with mechanisms similar to those proposed for venous infarction. Such cerebral parenchymal changes associated with DAVFs have been relatively rarely studied [5,6]. Although various imaging methods have been used to monitor cerebral changes associated with DAVF induced venous hypertentension, DWI findings have not been reported, to the best of our knowledge. Here we report on a DAVF case with diffuse white matter diffusion abnormality and T2 abnormality that reversed after treatment.

1. Case report

This 67-year-old female experienced a transient ischemic attack and a reversible ischemic neurologic deficit characterized with mild left hemiparesis within the last 6 months. This time she presented with mixed aphasia and subsequent loss of consciousness that had propagated within the last 24 h. T2 and fluid attenuated inversion recovery weighted magnetic resonance (MR) images showed prominent cortical veins and diffuse white matter hyperintensity in the temporal, parietal and occipital lobes (Fig. 1(a)). In some regions the cortical

* Corresponding author. Tel.: +90 232 4125913; fax: +90 232 2590541.
E-mail address: suleyman.men@deu.edu.tr (S. Men).

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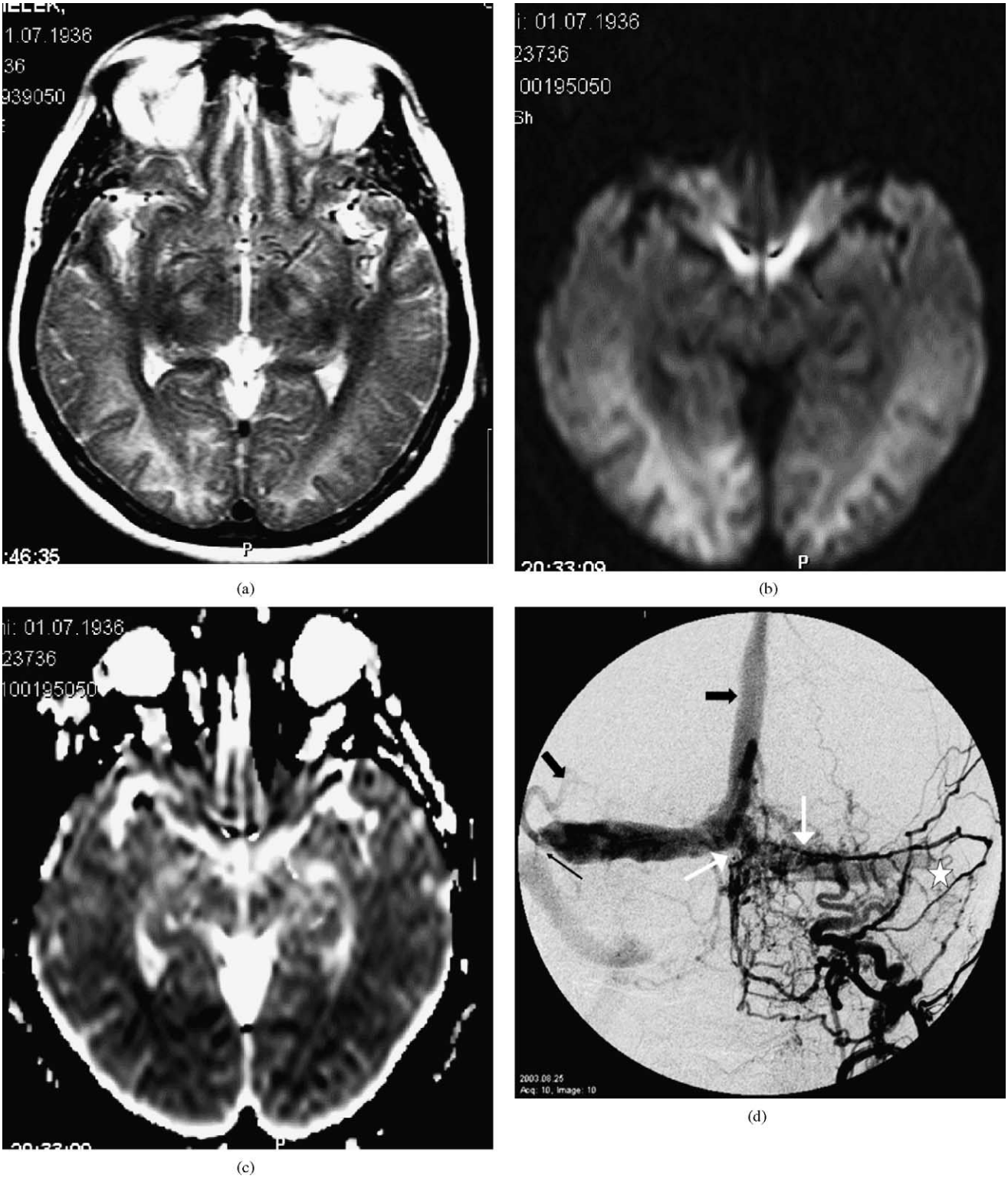

Fig. 1. (a) T2 weighted image. Diffuse white matter hyperintensity is seen in temporal and occipital lobes on both sides. (b, c) Diffusion weighted image and ADC map show restricted diffusion in the abnormal white matter areas. (d) Left external carotid angiogram shows the DAVF draining into torcular herophili and the left transvers sinus (white arrows). Note reflux (thick black arrows) into the superior sagittal sinus and cortical veins, occlusion of the left sigmoid sinus (white star) and severe stenosis of the right transverse sinus (thin black arrow). (e, f) Left carotid angiogram obtained in anterior–posterior (e) and lateral (f) projections after balloon angioplasty shows restored caliber of the right transverse sinus (arrow). The DAVF is still present and drains through the right transverse-sigmoid sinus, but there is no reflux into the sinuses or cortical veins. (g, h) Six months later both T2 (g) and diffusion (h) anormality reversed. Note the residual infarct (hollow arrow) and small chronic subdural hematoma from the previous cortical venous rupture.

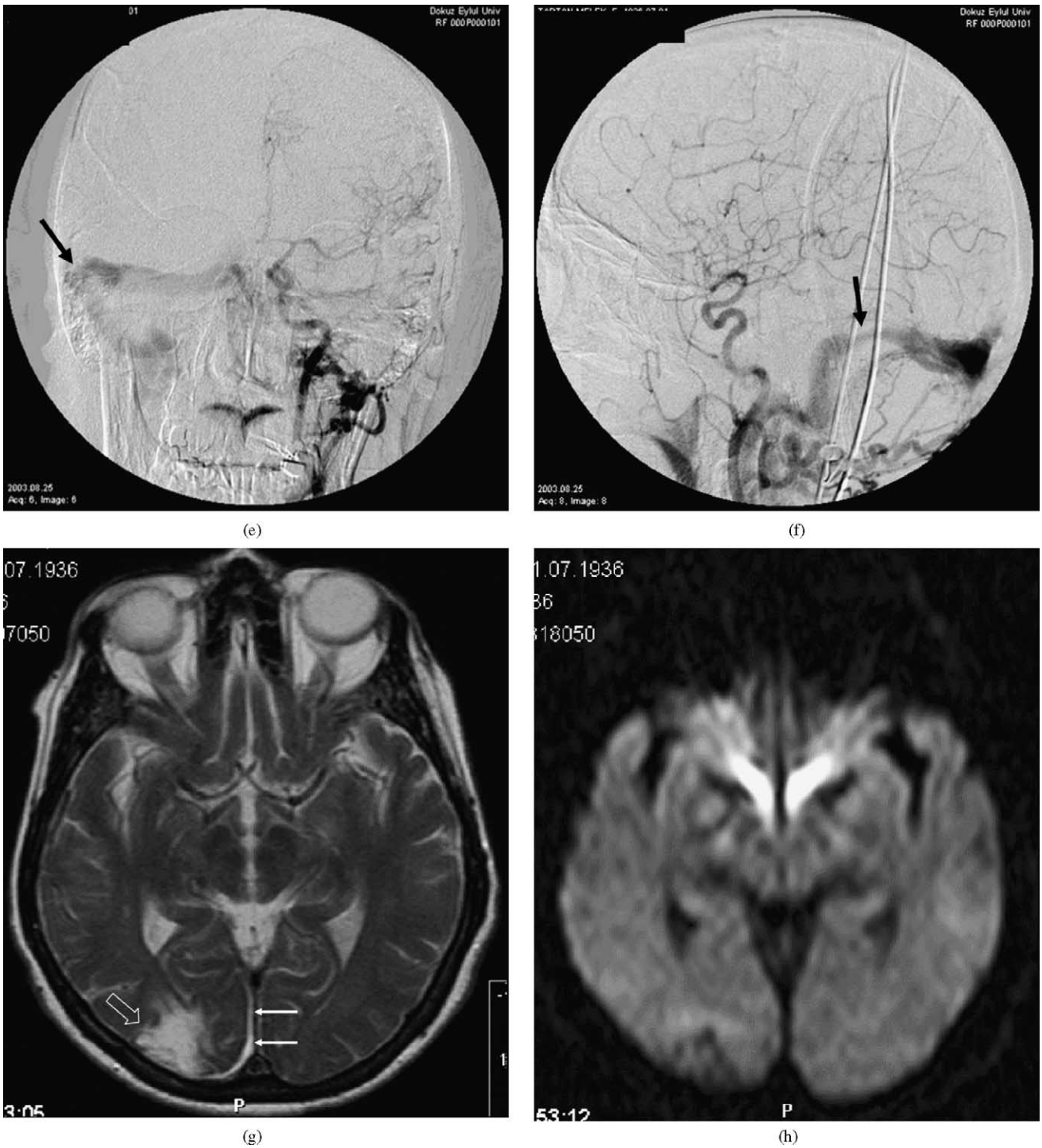

Fig. 1. (*Continued*).

gray matter was also affected; however, this was subtle. The hyperintense areas also showed restricted diffusion on the diffusion weighted imaging (Fig. 1(b, c)). MR arteriography, MR venography and the subsequent digital subtraction angiography confirmed DAVF of the torcular herofili, severe stenosis of the right transverse-sigmoid sinus junction, occlusion of the left sigmoid sinus and reversal of the flow in the superior sagittal and straight sinus (SSS) (Fig. 1(d)). The feeders were from the trans-osseous branches of the occipital artery on both sides. The SSS drained through anastomotic cortical veins. There was resistance to antegrade flow through the stenotic right sigmoid sinus evidenced by retrograde filling of the vein of Labbè on this side. Venous phase of the carotid angiography was delayed on both sides. Furthermore,

during the venous phase, no opacification of the venous sinuses could be shown. Instead, the venous return of the cerebral hemispheres was accomplished by the collateral venous network. The patient underwent balloon angioplasty of the stenotic right sigmoid sinus and subsequently transarterial particle embolization of the DAVF. These interventions eliminated venous outflow restriction and restored antegrade venous flow again (Fig. 1(e, f)). During the balloon angioplasty, the guide wire ruptured a cortical vein in the right occipital lobe. This resulted in hemorrhage and parenchymal damage in a focal area of the right occipital lobe and a small self-limiting parafalcine subdural hematoma (Fig. 1(g)). Clinically, the patient improved progressively, although she had a sequel visual defect on the left side. Six months later, she was free of neurologic deficits except some residual partial visual defect, and she was able to look after herself without anyone's help. The 6-month MR images showed reversal of both T2 hyperintensity and diffusion abnormality (Fig. 1(g, h)).

2. Discussion

In DAVFs with antegrade flow obstruction and reflux into superior sagittal sinus, straight sinus or cortical veins, the retrograde transmission of pressure results in intracranial venous hypertension. This venous hypertension and the resultant congestion in turn impair parenchymal venous drainage, thereby causing ischemia [5–7]. Cerebral hemodynamic changes associated with DAVFs were studied with various imaging modalities including positron emission tomography (PET) and MR perfusion imaging [6,8]. MR perfusion showed reduced cerebral blood flow, increased cerebral blood volume, increased mean transit time associated with the transverse-sigmoid sinus DAVFs [8]. PET also showed impaired cerebral perfusion characterized with reduced CBF and increased oxygen extraction fraction in cases of DAVF with retrograge venous drainage [6]. These data indicate that the cerebral perfusion is impaired in patients with DAVF.

In our case, we observed diffuse cytotoxic edema evidenced by low appearant diffusion coefficient (ADC). Although many imaging features of DAVFs have been described, no DAVF case with diffusion abnormality has been reported. DWI provides an early indicator of cerebral ischemic damage. Water shifts from the extracellular to the intracellular compartment (i.e., cytotoxic edema) that occur at the early stage of arterial stroke are thought to be responsible for the hyperintensity on DWI and for the drop in ADC values. DWI can reliably distinguish vasogenic from cytotoxic edema. While cytotoxic edema is characterized by restricted diffusion, vasogenic edema is characterized by elevated diffusion due to a relative increase in water in the extracellular compartment. On DWI, vasogenic edema may be hypointense to slightly hyperintense because these images have both T2 and diffusion weighting. When vasogenic edema is hyperintense on DWI, it can resemble hyperacute or subacute infarction. On ADC images, cytotoxic edema is hypointense and vasogenic edema is hyperintense.

DAVFs and venous thromboses have similarities. Both in cerebral venous thrombosis and DAVFs with pial venous reflux, the leading cause of the parenchymal damage is the venous hypertension. In approximately 50% of the cases, cerebral venous thrombosis results in vasogenic edema and infarction. The pathophysiology of venous infarction has been studied using various experimental models. In these models, raised intracranial pressure, increased cerebral blood volume, decreased cerebral blood flow and reduced cerebral perfusion pressure were found to be the consequences of venous outflow obstruction and resulted in venous infarction and hemorrhage [2–4]. In an experimental study on rats the major pathological event was the development of cytotoxic edema indicated by low ADC values within the first 1–2 h of venous occlusion. The magnetic resonance perfusion imaging in the same study revealed reduced perfusion in the same period suggesting decreased perfusion pressure and consecutively occurring energy depletion and membrane failure as the cause of cytotoxic edema (2). The observations in humans as reported in some studies did show early cytotoxic edema and subsequent vasogenic edema in some patients with venous infarction; however, the DWI pattern of human cerebral venous obstruction did not closely follow the results of animal studies [3,4,9,10,11]. First of all, DWI abnormality was not consistently shown in all patients; while some lesions in some patients showed pure cytotoxic edema, some others showed pure vasogenic edema or a mixture of both. Furthermore, some lesions contained hemorrhagic components, which further complicated diffusion imaging because of the vulnaribility of DWI with echo planar imaging to magnetic succeptibility effects of hemorhage. The follow-up imaging showed an increase in the ADC in all cases with initial diffusion restriction. In the long-term follow-up, however, some lesions with restricted diffusion evolved into persistent T2 abnormality while others healed without any radiological or clinical sequela. The discrepancy between the results of the experimental and the clinical studies likely results from the difference in the time course of venous thrombosis: in the laboratory animals the thrombosis is completed in a few minutes, whereas it is difficult to estimate the interval between the onset of thrombosis and the time at the diagnosis in humans [10].

Abnormal T2 hyperintensity in our case again suggests coexistence of vasogenic edema. The venous infarction models propose that retrograde venous pressure results in blood brain barrier disruption and consequently vasogenic edema and hemorrhage into extracellular space [2–4]. Although the abnormal T2 changes representing vasogenic edema do not appear before 4–6 h in the arterial stroke models, such changes appear much earlier in the venous stroke models [2]. This observation is not surprising regarding the coexistence of increased venous pressure with resultant decreased cerebral perfusion pressure. A remarkable finding in our case was the reversibility of the diffusion abnormality. Although reversal

of impaired perfusion after treatment has been observed in DAVFs in two studies, neither a case of diffusion abnormality nor its reversal associated with a DAVF has been reported previously [6,8]. In the arterial stroke in humans, nearly all lesions characterized by restricted diffusion progress to infarction [1]. However, such is not the case for the venous stroke [1,3]. The results of a previous study have shown that areas with decreased ADC values associated with cerebral venous thrombosis may be reversible [3]. As speculated in that study, the tissue affected by the venous stroke can still be perfused if there is sufficient collateral venous flow. With such flow conditions, swollen cells might be functionally but not irreversibly damaged, and therefore have a potential for recovery. The reversal of both cytotoxic and vasogenic edema after reconstitution of the antegrade venous flow in the present case supports the different nature of venous stroke from the arterial stroke.

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