Intracranial Haemorrhage Probably Due to an Angiographically Occult AVM after Carotid Stenting

A Case Report

M. BERKER, A. ULUS, S. PALAOGLU, F. SOYLEMEZOGLU, H. AY, S. CEKIRGE

Department of Neurosurgery, Hacettepe University School of Medicine; Ankara, Turkey

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Summary

Angiographically occult vascular malformations refer to cerebrovascular malformations that are not demonstrable on technically satisfactory cerebral angiography.

Authors herein present a very unusual intracranial bleeding complication related to an angiographically occult vascular malformation after extracranial carotid artery stenting procedure.

A 52-year-old male patient admitted to the hospital with 2 episodes of amaurosis fugax in the left eye. Cervical carotid angiography and bilateral carotid Doppler ultrasonography revealed a 98% stenosis of the left internal carotid artery just distal to the bifurcation. Post-stenting control cervical carotid angiography revealed neither any residual stenosis nor a developmental venous anomaly.

The patient developed left pupil dilatation with loss of consciousness two hours after the neurovascular intervention. Emergent cranial CT showed acute subdural haematoma, intracerebral and subarachnoid haemorrhage with massive midline shift. He underwent an emergent craniotomy with left temporal lobectomy. Abnormal cortical vascular structures with prominent engorgement were remarkable over the posterior temporal cortex. Histopathological studies confirmed the diagnosis of an occult AVM

Classically, these lesions are not visualized with angiography. Our patient's cerebral angiography and MR investigations were all normal. To our knowledge this is the first case in literature in which intracranial haemorrhage (acute subdural haematoma, intracerebral haematoma, SAH) occurred due to hyperperfusion of angiographically occult vascular malformation.

Introduction

Angiographically occult vascular malformation terminology was introduced by Crawford and Russell in 1956 to designate small vascular malformations, which escape angiographic demonstration but are identified histologically as causing spontaneous cerebral haemorrhage [1]. Later the term has been expanded to encompass all vascular malformations, which are not detected by angiography. Therefore it seems more appropriate to call this group of vascular malformations as "angiographically occult (or cryptic) cerebral vascular malformations" [2]. However, it is very important to obtain technically satisfactory cerebral angiography and magnetic resonance imaging (MRI) before naming these lesions as angiographically occult vascular malformation [3].

Intracranial haemorrhage resulting from hyperperfusion syndrome is a well-known com-

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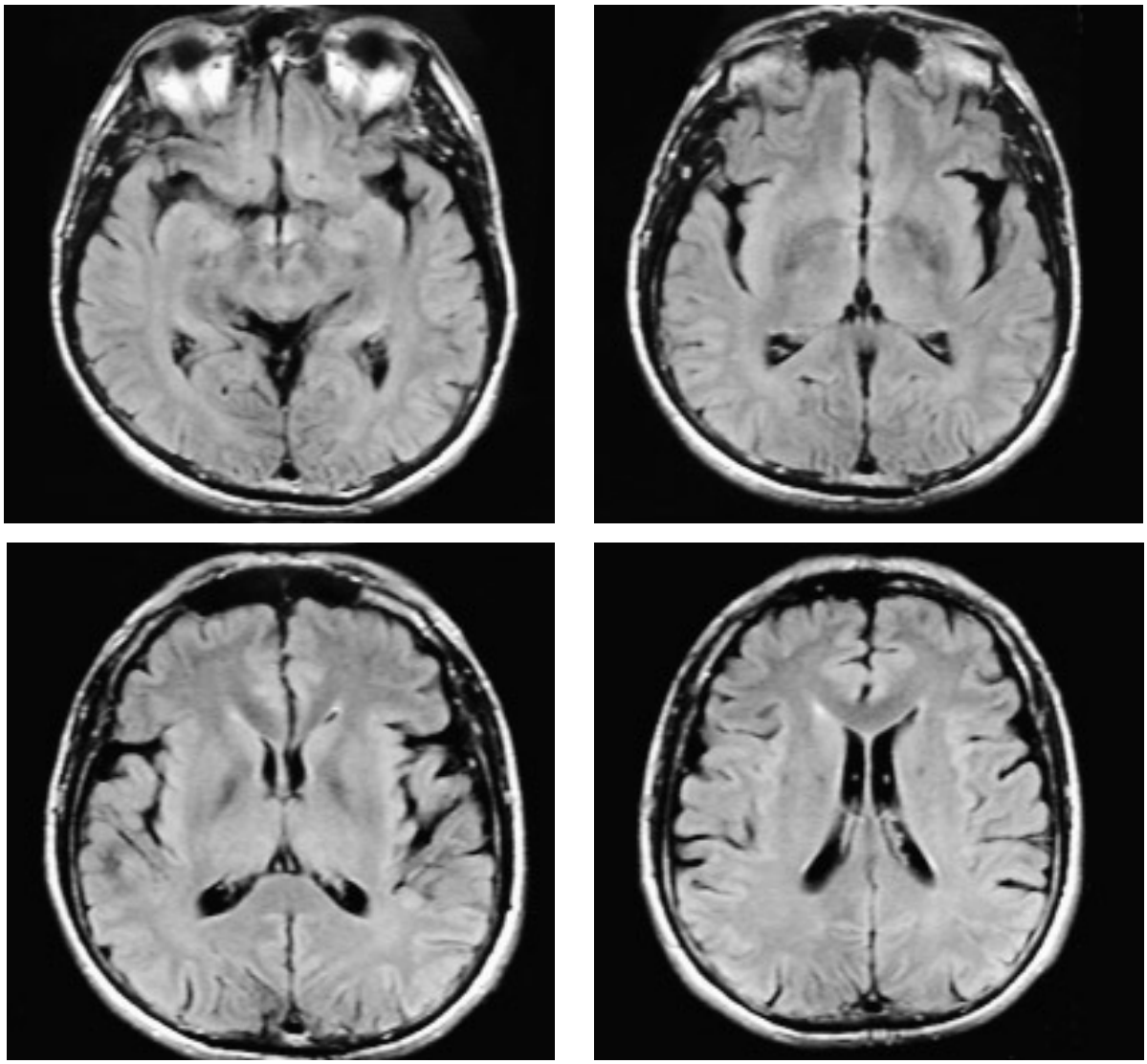

plication of carotid artery angioplasty, stent placement and carotid endarterectomy procedures [4,5,6]. Intracranial haemorrhage due to angiographically occult AVM after carotid stenting procedure has not been reported to our knowledge. Here, we present intracranial bleeding after extracranial carotid artery stenting with a very unusual probable cause i.e. angiographically occult AVM.

Case Report

A 52-year-old male patient was admitted to the hospital with 2 episodes of amaurosis fugax in the left eye. Neurological examination was normal. Before the endovascular treatment, non contrast (NCECT) and contrast enhanced (CECT) cranial CT, cranial MRI including diffusion weighted sequence and MRA of cervical vessels were obtained. Angiographic work-up consisted of aortic arch, bilateral common carotid and bilateral vertebral arteries including intracranial vasculature.

NCECT and CECT, cerebral angiography, and brain MRI studies were normal. Cervical carotid angiography and bilateral carotid color Doppler ultrasonography revealed a 98% stenosis of the left internal carotid artery just distal to the bifurcation. The patient was put on aspirin (300 mg) and clopidogrel (75 mg) treatment. Carotid artery stenting was performed a week later. Neither pre stenting MRI nor the

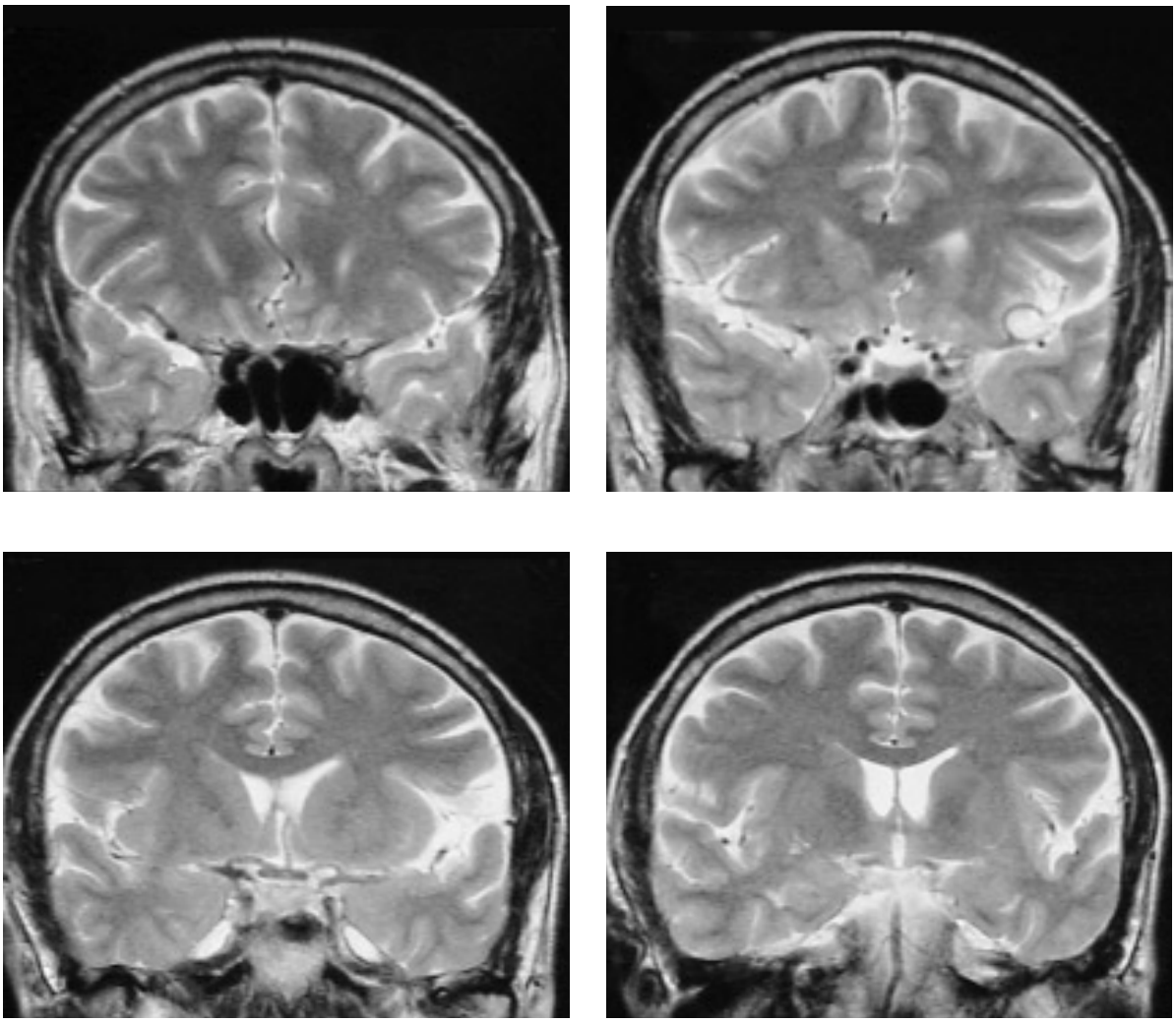

Figure 1 Pre-stenting cranial MRI: Transverse FLAIR (A) and T2 coronal (B) subsequent images do not show any evidence of arteriovenous malformation.

pre and post stenting angiography reveal any cerebral arterial and/or venous abnormality (figure 1,2). Post-stenting control cervical carotid angiography revealed no residual stenosis and no developmental venous anomaly was noted in this study. Immediately before the procedure, the patient was given heparin at a bolus dose of 2000 units followed by continuous intravenous infusion of 500 U/hour. Moreover, one mg tirofiban was administered as a 30-minute intravenous infusion starting just before the stent deployment; in order to prevent procedure related embolic complications, as a part of our stent protocol. The patient had severe headache and nausea an hour after the intervention. Two hours later, he developed left pupil dilatation with loss of consciousness. His INR and aPTT were checked to exclude the possibility of a haemorrhagic complication secondary to anticoagulation and his aPTT and INR were found slightly elevated (aPTT: 49.7, INR: 1.34). On cranial CT, there was acute intracerebral, subarachnoidal and subdural haemorrhage associated with marked midline shift. The haemorrhage was in the left temporal region but extends to frontal and parietal. Intracerebral component was approximately 4x5 cm and there existed consequent edema around (figure 3). Pupils were dilated with no response to light. Emergent left temporal craniotomy and haematoma evacuation were performed. His neurological deterioration slightly im-

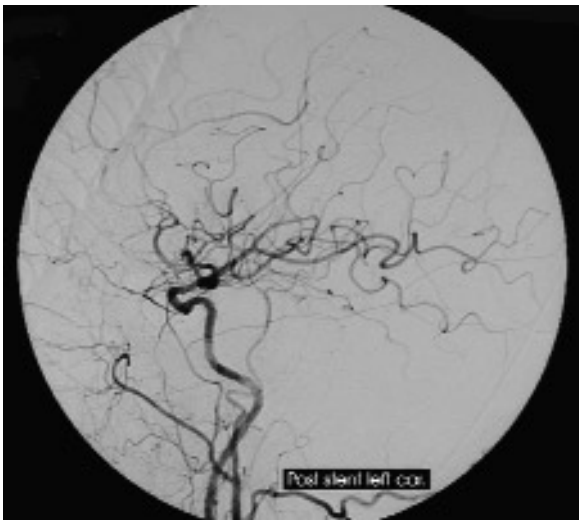

Figure 2 Post- stenting lateral carotid angiogram is normal with no evidence of abnormal vasculature or any arterial occlusion.

proved in the early postoperative period with significant left-sided anisocoria and extensor extremity response. The follow up cranial CT scan showed significant oedema and residual subdural haematoma in the left temporal region 24 hours after the surgery. He underwent a second surgery with left temporal lobectomy. Abnormal cortical vascular structures with prominent engorgements were remarkable over the posterior temporal cortex.

Histopathological studies confirmed the diagnosis of an occult AVM. He did not respond to intensive medical treatment and died 6 days after the second surgery.

Discussion

"Angiographically occult vascular malformations" (AOVMs) of the brain and spinal cord represent a heterogeneous subset of lesions including cryptic arteriovenous malformations, cavernous malformations, capillary telangiectasias, and mixed lesions. AOVMs account for 8 to 15 % of intracranial vascular malformations. Classically, these lesions are not visualized with angiography [7]. Our patient's cerebral angiography and MR investigations were all normal.

In our case there was high-grade stenosis (98%) of ICA. Due to loss of autoregulation, hyperperfusion might have occurred in the ipsilateral ICA territory after stenting and the angiographically occult AVM caused massive subdural and subarachnoid haemorrhage.

Histopathological examination of the specimens showed severe intraparenchymal and subarachnoidal haemorrhage. Acute ischemic changes and edema were prominent in the cortices. The histopathological examination of the macroscopically small abnormal vascular lesion revealed an obvious increase in both the number and size of the blood vessels.

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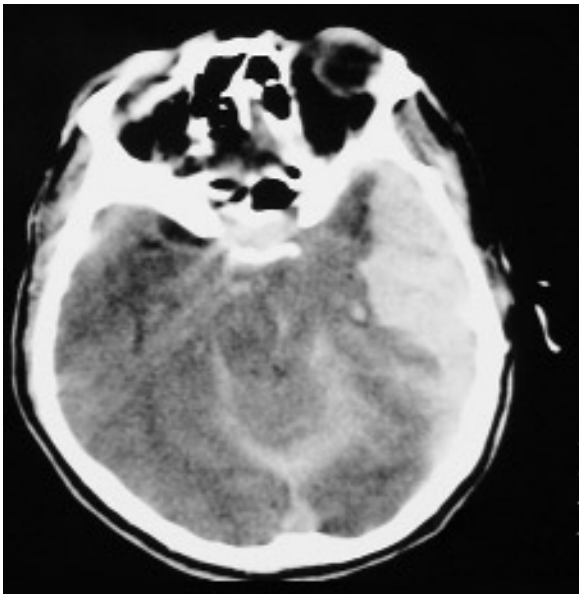

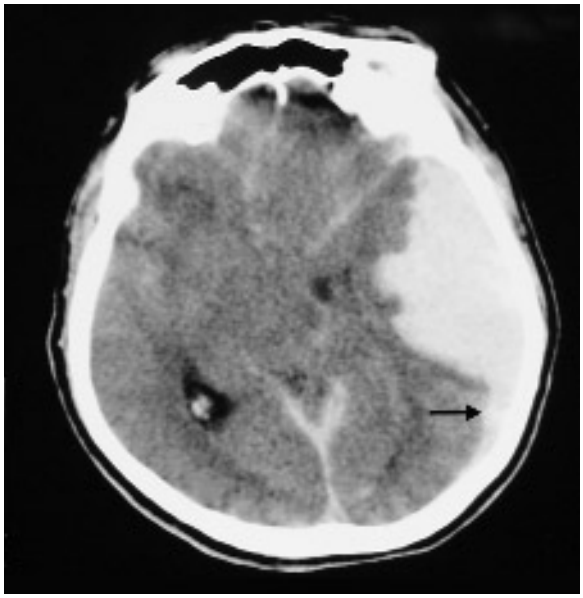

B

Figure 4 Cranial CT: Non contrast CT showed massive intraparenchymal and extra-axial acute haemorrhage with edema, mass effect, midline shift, uncal and infratentorial herniation. Note the crescent shape of the extraaxial haemorrhage (arrow).

This small vascular malformation was located underneath the thickened leptomeninges in the subarachnoidal area, and abnormally sized vessels were penetrating the adjacent cortex. The abnormal vessels, as demonstrated by EVG (Elastica von Gieson) staining, were dilated arterial and venous structures, but there was no apparent mural abnormality. These findings were consistent with the morphological features of a vascular malformation, more specifically an arteriovenous malformation, located in the superficial cortex and subarachnoid space.

To our knowledge this is the first case in literature in which intracranial haemorrhage (acute subdural haematoma, intracerebral haematoma, SAH) occurred probably due to hyperperfusion of angiographically occult AVM.

Although cerebral hyperperfusion syndrome is a well-documented complication of carotid artery angioplasty and stent placement, it was originally described after surgical endarterectomy procedures with a reported incidence of 0.3 to 1.2% [5]. The incidence after cerebral angioplasty and stenting is unknown. Spetzler et al. described hyperperfusion which occurs after surgical resection of AVM's, due to loss of autoregulation in the surrounding normal brain parenchyma causing intracerebral haemorrhage [8].

In 1981, Sundt et Al described a triad of complications after carotid endarterectomy, including atypical migrainous phenomena, transient focal seizure activity and intracerebral haemorrhage. They used "hyperperfusion syndrome" term to describe this triad [9].

The triad was not complete in all patients. Cerebral hyperemia may cause headache, vomiting, confusion, arterial hypertension, focal neurological deficits, seizures, and subarachnoid haemorrhage after revascularization. It has been subsequently reported that there was also significant risk of hyperperfusion in other cerebral revascularization techniques such as extracranial carotid angioplasty and stent placement. Surgical risk factors for hyperperfusion are symptomatic high-grade stenosis, significant pressure gradient across the stenosis with poor distal perfusion of the ipsilateral hemisphere, poor cerebral collateral blood flow, contralateral carotid occlusion, perioperative hypertension, and the use of anticoagulant and antiplatelet agents.

Stenting of the carotid arteries has emerged as a potential treatment alternative for symptomatic and asymptomatic carotid stenosis.

The major concern regarding this form of therapy has been the risk of thromboembolism. To minimize the risk of stroke, pharmacological and mechanical approaches are currently being investigated. Considering the success of GP IIb/IIIa inhibitors in reducing ischemic complications during coronary interventions, these agents have been increasingly used in carotid stenting. However, there is substantial concern that risk of intracranial haemorrhage may be increased with GP IIb/IIIa inhibition 10. More than 80% stenosis is more likely to develop hyperperfusion and 90 to 99% stenosis carries even greater risk of hyperperfusion development.

Embolic complications are the major clinical case at in the carotid procedures that usually occur at the time of balloon inflation or stent placement. Therefore, we use GPIIb/IIIa inhibitors only during the procedure. So, a lower dose protocol we used was one forth of that routinely used in coronary procedures and it might be justified by rendering the risk of haemorrhage to lower rates. Reperfusion of an ischemic hemisphere may lead to a hyperperfusion syndrome in carotid artery procedures. This usually occurs after a delay of 24-48 hours and the antiplatelet medication may certainly increase the risk of haemorrhage.

However, in the presence of an occult AVM, haemorrhages might appear at an earlier time. In a patient with severe carotid stenosis, not only hypertension to provide the normal perfusion, but also ischemia due to the hypoperfusion may lead the loss of autoregulation. After the reperfusion, impaired autoregulation could be cause to the appearence and bleeding of the occult AVM's

In conclusion, authors report their anecdotal experience in a patient presenting with fatal bleeding following extracranial carotid stenting probably caused by an 'occult' AVM and, hence, would like to raise a question about the cause of poststenting reperfusion bleedings.

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Mustafa Berker, M.D.
Hacettepe University School of Medicine
Department of Neurosurgery
Altindag, 06100 - Ankara Turkey
E-mail: mberkermd@yahoo.com
mberker@hacettepe.edu.tr