



DOI: 10.5137/1019-5149.JTN.16613-15.2

Received: 28.12.2015 / Accepted: 20.4.2016

Published Online: 09.08.2016

Case Report

Bilateral Thalamic Edema from Coexisting Choroid Plexus Arteriovenous Malformation and Sinus Thrombosis: Case Report

Ryan B. KOCHANSKI¹, Andrew K. JOHNSON², Roham MOFTAKHAR³

¹Rush University, Medical Center, Department of Neurosurgery, Chicago, IL, USA

²Wellstar Medical Group, Department of Neurosurgery, Marietta, Georgia, USA

³Palmetto Health Neurosurgery Associates, Columbia, SC, USA

ABSTRACT

Bilateral thalamic dysfunction secondary to venous congestion may result from either venous sinus thrombosis or high flow arteriovenous malformations or a combination of both. We present a case of bilateral thalamic edema resulting from concomitant choroid plexus arteriovenous malformation (AVM) and straight sinus thrombosis and describe our treatment approach. The patient presented with several weeks of progressive confusion and memory deficits. Magnetic resonance imaging and venography (MRI/MRV) showed bilateral thalamic T2 hyperintensities and straight sinus thrombosis. Subsequent cerebral angiography revealed a choroid plexus AVM within the right lateral ventricle. The patient underwent surgical resection of the AVM resulting in postoperative resolution of bilateral thalamic edema on MRI and improvement of his confusion and memory deficits. This case demonstrates a rare example of reversible bilateral thalamic edema secondary to venous hypertension from both an AVM and sinus occlusion after appropriate treatment of the AVM.

KEYWORDS: Choroid plexus, Arteriovenous malformation, Sinus thrombosis, Thalamic edema

INTRODUCTION

Bilateral thalamic edema from venous hypertension may result from deep venous sinus thrombosis or high-flow arteriovenous malformations/fistulae due to arterialized venous outflow (1,4,6,8,12,15). Patients with thalamic dysfunction often present with dementia-like symptoms including cognitive decline, hypersomnolence and abulia (1,4,6,8,12,15).

Here we present a rare case of thalamic dementia caused by bilateral thalamic venous hypertension secondary to an underlying choroid plexus arteriovenous malformation (AVM) with superimposed straight sinus thrombosis. Choroid plexus AVMs are rare entities and present most frequently with intraventricular hemorrhage due to rupture (1, 3–9,11–13, 15, 16).

We report a unique case of symptomatic bilateral thalamic edema caused by both a choroid plexus AVM and thrombosis of the deep venous system. This combination of lesions presented unique challenges and considerations in the treatment approach, particularly in deciding whether to treat the sinus thrombosis, the AVM or both.

CASE REPORT

A 58 year-old, previously healthy male presented had a two-week history of confusion, disorientation and difficulty concentrating. On exam, his only neurologic deficit was poor memory recall. Brain magnetic resonance imaging (MRI) revealed bilateral T2-weighted thalamic hyperintensities (Figure 1A) as well as increased vascularity within the atrium of the right lateral ventricle. Magnetic resonance venography (MRV)



Corresponding author: Ryan B. KOCHANSKI

E-mail: Ryan_B_Kochanski@rush.edu

was concerning for straight sinus thrombosis (Figure 1B). Cerebral digital subtraction angiography (DSA) was performed to better characterize the MRI and MRV findings which demonstrated a Spetzler-Martin grade II choroid plexus AVM located within the atrium of the right lateral ventricle with feeding vessels from the postero-lateral choroidal arteries and draining vein emptying into an engorged basal vein of Rosenthal near the anterior choroidal point (Figure 1C). Furthermore, it was noted on the angiogram that the straight sinus was occluded.

Given the presence of both the choroid plexus AVM and straight sinus thrombosis, the decision was made to treat the AVM initially. Preoperative embolization was not feasible due to the small feeding arteries of the postero-lateral choroidal arteries. With these factors taken into account, it was decided to perform microsurgical excision of the AVM via a right superior parietal lobule approach. An endoscope was initially placed through the superior parietal lobule for visualization of the AVM prior to resection (Figure 2). The AVM was subsequently resected using microsurgical techniques. Intraoperative, post-resection cerebral angiography demonstrated complete obliteration of the AVM (Figure 3A). The patient had an uneventful post-operative course and was eventually discharged to an acute rehabilitation facility. MRI at 3-month follow-up showed resolution of bilateral thalamic T2 weighted

hyperintensities (Figure 3B). The patient had marked improvement in his memory and cognition allowing him to return to functional independence.

DISCUSSION

Previously described cases of thalamic dementia have demonstrated reversal of thalamic edema on MRI and resolution of symptoms once the source of venous congestion has been treated (1,4-7,9,10). In the setting of sinus occlusion, anticoagulation is considered the primary treatment for sinus thrombosis, even in the setting of hemorrhage, and revascularization is sometimes considered (3). In our case, medical therapy alone with anticoagulation was not considered because of the inherent risk of AVM rupture and the likelihood that symptoms would not improve without treating the AVM. Moreover, it has been postulated by Viñuela et al. that the mechanism of sinus occlusion in patients with concomitant arteriovenous malformations may be secondary to local endothelial damage caused by turbulent or unstable hemodynamic flow caused by the pre-existing AVM (17). Radiosurgery has also been described for treatment of choroid plexus AVMs but was not performed because of the significant edema already present within the thalami (13). Moreover, several cases of occlusive

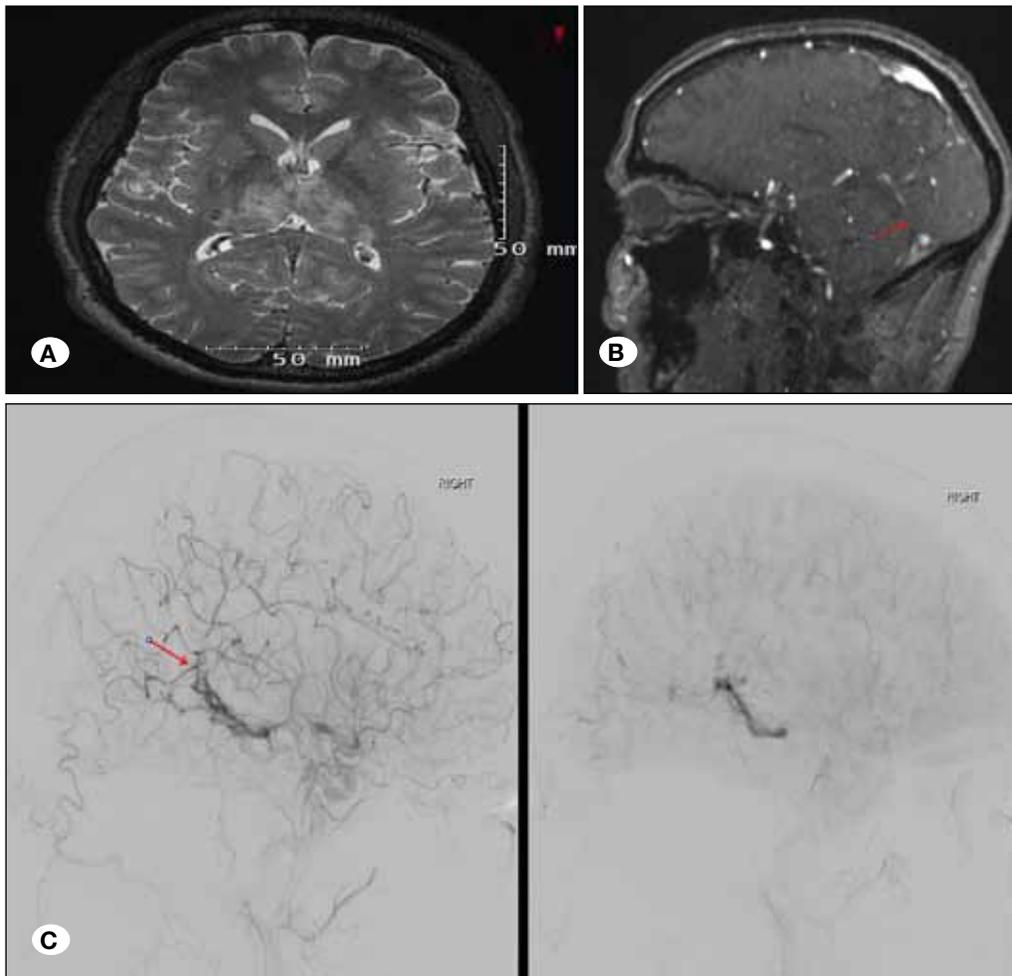


Figure 1: **A)** Preoperative T2-weighted axial MRI demonstrating bilateral thalamic hyperintensities. **B)** Preoperative sagittal MRV demonstrating occlusion of the straight sinus. **C)** Preoperative lateral projection DSA of the right internal carotid artery demonstrating the AVM nidus (arrow) with feeding vessels from the postero-lateral choroidal arteries and draining vein into the basal vein of Rosenthal.

hyperemia caused by impaired venous drainage following AVM radiosurgery have been described, and in these reports, the authors hypothesize that the clinical deterioration of the reported patients was likely secondary to local hemodynamic changes leading to premature thrombosis of draining veins (2,14). Given the pre-existing straight sinus thrombosis and thalamic edema already present in our patient, radiosurgery was ultimately not considered as it could potentially further worsen thrombosis of the deep venous system.

The presented patient likely had longstanding, asymptomatic venous hypertension from the choroid plexus AVM that became symptomatic after thrombosis of the straight sinus. A prior case of de novo sinus thrombosis distal to an arteriovenous fistula (AVF) has been described, also supporting high-flow occlusive venopathy as an etiology for sinus throm-

bosis in this setting (16). In that case, both the AVF and sinus thrombosis were treated, but in our case, clinical improvement was achieved with treatment of the AVM only. Microsurgical excision of the AVM was pursued, as it seemed to be the safest way of reducing both venous congestion and the risk of AVM hemorrhage by addressing the causative lesion. This treatment strategy was also thought to obviate the need for anticoagulation at present and in the future under the assumption that reduction of the turbulent venous hemodynamics through resection the AVM may lead to reduction of venous endothelial damage and spontaneous resolution of thrombosis. Although venous imaging was not performed at follow-up to evaluate the straight sinus, the thrombosis was assumed to be resolved/improved given the complete absence of thalamic edema on the follow-up MRI and the marked improvement in the patient's symptoms. Consistent with the above described hypothesis by Viñuela et al., we assume that the reduction in venous hypertension after AVM resection likely mitigated the turbulent hemodynamics within the deep venous system thus allowing for spontaneous recanalization of the straight sinus (17). Had the patient failed to improve clinically or if thalamic edema persisted on MRI, further investigation of a potential persistent sinus occlusion would have been pursued and treated appropriately.

■ CONCLUSION

Symptoms from bilateral thalamic edema caused by increased venous hypertension can be reversed with appropriate management of the inciting vascular lesion. This case demonstrates a rare example of concurrent AVM and sinus occlusion, emphasizing the importance of careful angiographic evaluation in the setting of sinus occlusion. The case adds support for a mechanism of sinus occlusion caused by high-flow arteriovenous lesions.

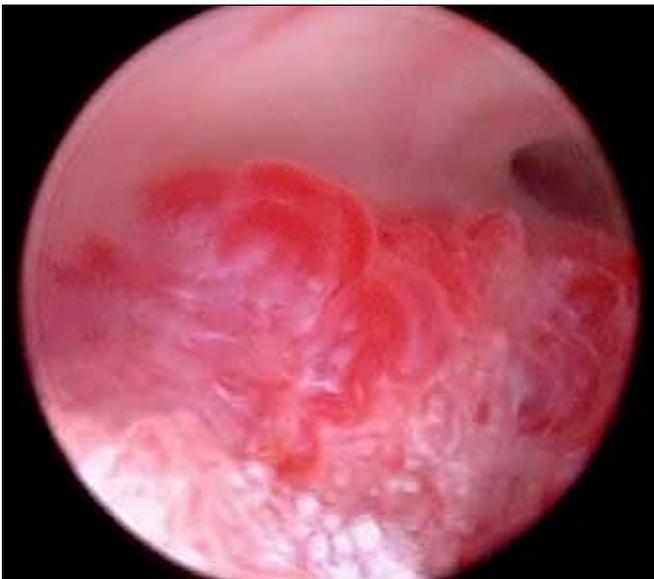


Figure 2: Intraoperative endoscopic view of the AVM.

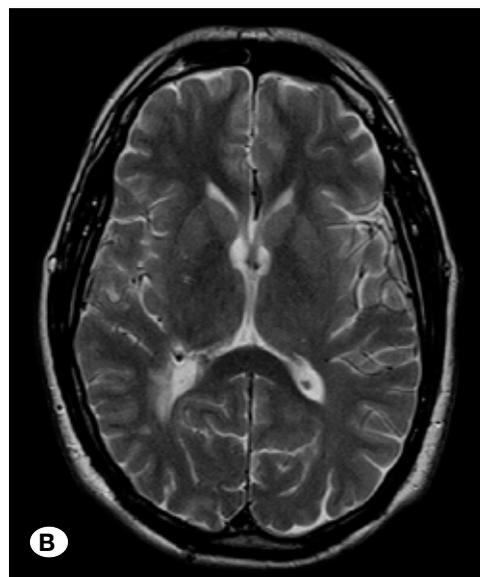
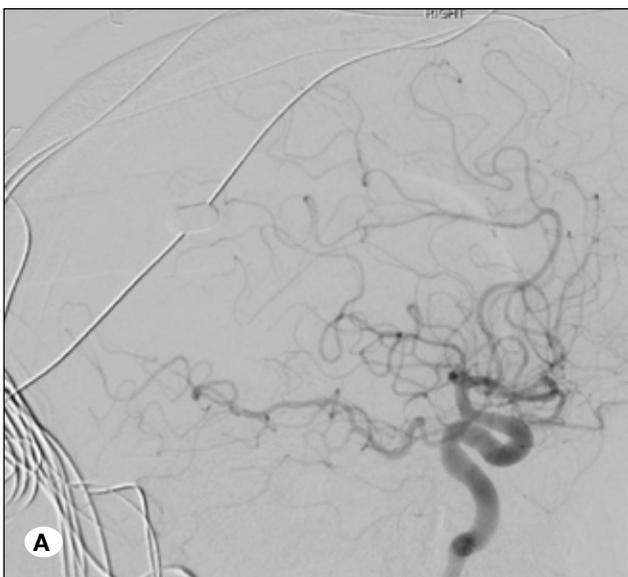


Figure 3:
A) Intraoperative lateral projection DSA of the right internal carotid artery showing complete resection of the AVM.
B) Three month postoperative T2-weighted axial MRI showing resolution of bilateral thalamic hyperintensities.

■ REFERENCES

- Bezerra DC, Michel P, Maulaz AB, Binaghi S, Bogousslavsky J: Resolution of bilateral thalamic lesions due to deep cerebral venous thrombosis. *Arch Neurol* 62:1638–1639, 2005
- Chapman PH, Ogilvy CS, Loeffler JS: The relationship between occlusive hyperemia and complications associated with the radiosurgical treatment of arteriovenous malformations: Report of two cases. *Neurosurgery* 55:228–233; discussion 233–234, 2004
- Coutinho J, de Bruijn SF, Deveber G, Stam J: Anticoagulation for cerebral venous sinus thrombosis. *Cochrane Database Syst Rev* 8:CD002005, 2011
- Goncalves MB, Maia Jr O, Correa JLA, Siqueira SBP de, Christoph D de H, Landeiro JA: Dural arteriovenous fistula presenting as thalamic dementia. *Arq Neuropsiquiatr* 66: 264–267, 2008
- Greenough GP, Mamourian A, Harbaugh RE: Venous hypertension associated with a posterior fossa dural arteriovenous fistula: Another cause of bithalamic lesions on MR images. *AJNR Am J Neuroradiol* 20: 145–147, 1999
- Gupta R, Miyachi S, Matsubara N, Izumi T, Naito T, Haraguchi K, Wakabayashi T: Pial arteriovenous fistula as a cause of bilateral thalamic hyperintensities--an unusual case report and review of the literature. *J Neurol Surg Part A-Cent Eur Neurosurg* 74:e18–24, 2013
- Holder CA, Bell DA, Lundell AL, Ulmer JL, Glazier SS: Isolated straight sinus and deep cerebral venous thrombosis: Successful treatment with local infusion of urokinase. Case report. *J Neurosurg* 86:704–707, 1997
- Ito M, Sonokawa T, Mishina H, Sato K: Reversible dural arteriovenous malformation-induced venous ischemia as a cause of dementia: Treatment by surgical occlusion of draining dural sinus: Case report. *Neurosurgery* 37: 1187–1191; discussion 1191–1192, 1995
- Iwasawa E, Ishibashi S, Miki K, Yoshino Y, Nemoto S, Mizusawa H: Teaching neurolimages: Reversible cognitive impairment with bithalamic lesions caused by a dural arteriovenous fistula. *Neurology* 81:e38–39, 2013
- Matsumura A, Oda M, Hozuki T, Imai T, Shimohama S: Dural arteriovenous fistula in a case of dementia with bithalamic MR lesions. *Neurology* 71:1553, 2008
- Miyasaka Y, Yada K, Ohwada T, Morii S, Kitahara T, Kurata A, Tanaka R: Choroid plexus arteriovenous malformations. *Neurol Med Chir* 32: 201–206, 1992
- Morparia N, Miller G, Rabinstein A, Lanzino G, Kumar N: Cognitive decline and hypersomnolence: Thalamic manifestations of a tentorial dural arteriovenous fistula (dAVF). *Neurocrit Care* 17: 429–433, 2012
- Nataf F, Meder JF, Oppenheim C, Merienne L, Schlienger M: Radiosurgery of choroidal and cisternal cerebral arteriovenous malformations. *Neurochirurgie* 47: 283–290, 2001
- Pollock BE: Occlusive hyperemia: A radiosurgical phenomenon? *Neurosurgery* 47: 1178–1182; discussion 1182–1184, 2000
- Santillan A, Safdieh JE, Gobin YP, Patsalides A: Neurological picture. Bilateral thalamic venous hypertension caused by a tentorial dural arteriovenous fistula: Endovascular treatment. *J Neurol Neurosurg Psychiatry* 82: 749–750, 2011
- Song JK, Patel AB, Duckwiler GR, Gobin YP, Jahan R, Martin NA, Cacayorin ED, Viñuela F: Adult pial arteriovenous fistula and superior sagittal sinus stenosis: Angiographic evidence for high-flow venopathy at an atypical location. Case report. *J Neurosurg* 96:792–795, 2002
- Viñuela F, Nombela L, Roach MR, Fox AJ, Pelz DM: Stenotic and occlusive disease of the venous drainage system of deep brain AVM's. *J Neurosurg* 63:180–184, 1985