

Clinical Article

Surgical disconnection of cortical venous reflux as a treatment for Borden type II dural arteriovenous fistulae

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Summary

Objective. The presence of cortical venous reflux is recognized as an indicator of increased risk of intracranial hemorrhage and neurological deficits in cranial dural arteriovenous fistulas. Its disconnection is well accepted as a treatment for fistulas with direct cortical reflux (Borden type III), but the role of disconnection of the cortical venous reflux in the management of fistulas that involve the venous sinus and cortical venous reflux (Borden type II) is still a matter of debate. We analyze the experience of the Toronto Brain Vascular Malformation Study Group in the management of these lesions by simple cortical venous reflux disconnection and its impact in the future risk of bleeding.

Methods. From June 1984 to August 2004, 347 patients with dural arteriovenous fistulas, either cranial or spinal, were evaluated by the group. Fifty-three patients had a Borden type II dural arteriovenous fistulas. Twenty-five patients were submitted to simple surgical disconnection of the dural arteriovenous fistulas, two were lost for follow-up. There were 15 females and 8 males, with mean age at diagnosis of 53.9 years. Follow-up time was 112.6 patient-years, from 2 months to 11 years, mean 4.9 years. Endovascular treatment was attempted in all patients, but no disconnection was possible. Twelve

patients had their fistulas completely occluded by endovascular means, but are not analyzed here. There were four complications from the 93 endovascular procedures, and 3 from the 27 surgical procedures. Two patients required a repeated surgical procedure. No episode of intracranial hemorrhage or worsening neurological deficit was seen after disconnection of the cortical venous reflux in 4.9 years of follow-up.

Conclusion. Simple surgical disconnection of the cortical venous reflux maybe an option in the management of patients with Borden type II dural arteriovenous fistulas. This procedure is a much smaller surgical undertaking and is associated with fewer complications than attempts to resect or pack the whole fistula, especially if located in the skull base.

Keywords: Brain hemorrhage; cortical venous reflux; dural arteriovenous fistulae.

Introduction

Dural arteriovenous fistulas are uncommon lesions, comprising about 10%–15% of all intracranial arteriovenous malformations. Thought to be acquired, these lesions consist of one or more anomalous arteriovenous connections located in the dura-mater, usually close or in the walls of a cranial venous sinus. More commonly diagnosed in the adult population, they may manifest with a variety of symptoms, including benign ones such

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as headache, bruit, ocular symptoms (exophthalmos, congestion, and ophthalmoplegia) or potentially devastating ones, such as intracranial hemorrhage and progressive cognitive symptoms as a result of venous congestion [13, 15, 20]. The knowledge on cranial arteriovenous dural fistulas is increasing progressively over the last decades. It's now well accepted that the presence of cortical venous reflux is an important risk factor for hemorrhage. Dural fistulas without cortical venous reflux usually have a benign clinical course, the opposite being true for the ones with this feature, which have a high yearly risk of hemorrhage [2–4, 6, 18, 20–23]. The conservative management of Borden type I and the venous disconnection (by surgical or endovascular means) of Borden type III fistulae are relatively well established treatment modalities [4, 5, 16, 18, 21, 23]. The proper management of Borden type II fistulae remains debatable. Considering the grim natural history of lesions with cortical venous reflux, with an annual mortality of 10.4% and an event rate (hemorrhagic and non-hemorrhagic neurological deficit) of 15%/year, it is clear that treatment should be offered to all patients. Because of the associated sinus involvement, disconnection of the cortical venous reflux only is not considered the best choice of treatment by many, since it disconnects the venous reflux but does not deal directly with the fistula itself. Surgical attempts to remove the involved sinus along with disconnection of the cortical venous reflux frequently involve complicated surgical procedures associated with significant blood loss, and endovascular occlusion of the whole fistula is not always a feasible option [7–11, 17, 19]. The Toronto Brain Vascular Malformation Study Group published in 2004 a series of patients with aggressive dural arteriovenous fistulas (Borden types II and III) where the results of the cortical venous reflux disconnection for part of the group were evaluated, with good outcomes [23]. We now expand that analysis, focusing on the group of patients with Borden type II fistulae submitted to the simple disconnection of the cortical venous reflux.

Material and methods

Since 1984 the Toronto Brain Vascular Malformation Study Group is collecting information on patients with vascular diseases of the central nervous system. From June 1984 to August 2004, 347 patients with diagnosed dural arteriovenous fistulas, either cranial or spinal, were evaluated by the group. All patients had at least one complete angiogram, including external carotid artery

branches. Of those, 53 patients were diagnosed as having a dural arteriovenous fistula classified as a Borden type II fistula. Twenty-five patients were submitted to disconnection of the cortical venous reflux and 12 patients were “cured”, meaning either complete occlusion or resection of the fistula. Nine patients had no treatment and 7 had partial treatment, mainly because of refusal by the patients in starting or continuing with the suggested treatment plan. The subgroup of patients submitted to simple disconnection will be the object of this paper.

All patients were evaluated in a clinic attended by cerebrovascular neurosurgeons and interventional neuroradiologists, their management being discussed in a multidisciplinary conference. Treatment was offered to all patients with a Borden type II dural arteriovenous fistulas. The disconnection of the cortical venous reflux, consisting of simple interruption of the cortical venous drainage at its origin in the dura-mater, either by endovascular or open surgical means, with no attempts to deal with the intradural fistulous connections (Fig. 1) was the main goal of the treatment. Complete occlusion of the fistula was performed in occasions when clearly feasible and never at the cost of increased risk. All patients had at least one postoperative angiogram to document the complete absence of cortical venous reflux, or occlusion of the fistula, and when this was not the case, further treatment was offered. After that, patients were followed clinically and with non-invasive imaging by the group. Any change in the symptoms, including improvement (bruits, ocular symptoms) would prompt reevaluation with angiography.

Follow-up information was obtained at regular clinic visits or with telephone calls. In a subgroup of 9 patients was possible to obtain a late MRI/MRA study in order to evaluate the presence or not of cortical venous reflux. We evaluate here the outcome of the group of patients submitted to simple disconnection of the cortical venous reflux, mainly regarding incidence of intracranial hemorrhage after the treatment.

Results

Of the 25 patients submitted to cortical venous reflux disconnection, 2 were lost to follow-up (92% of follow-up). There were 15 females and 8 males. The mean age at diagnosis was 53.9 years, ranging from 29 to 79 years. The follow-up time was 112.6 patient-years, from 2 months to 11 years, mean 4.9 years. The most common location of the fistula was the cavernous sinus (17 patients), followed by the transverse sinus (4 patients), the

torcular region and superior sagittal sinus (1 patient each). Accordingly, ocular symptoms predominate at presentation – 17 patients, 16 with CS fistulae and 1 with a TS fistula. Hemorrhage was the presenting symptom in 2 patients, both with fistulae located at the TS, tinnitus in 2 patients (1 in the CS, 1 in the TS), headache in one patient (TS) and cognitive symptoms in another (SSS).

Endovascular treatment was performed in all patients, either aiming to convert an aggressive Borden type II in a benign Type I fistula, at complete occlusion or at palliation of symptoms. The pre-treatment goal was to cure the dural arteriovenous fistulas in 14 patients (disconnection of the cortical venous reflux being here accepted as “cure”) and palliative (symptomatic) treatment in 9 patients. Ninety-three endovascular procedures were performed, ranging from 1 to 6 procedures per patient. Complete occlusion of the fistula was possible in twelve cases. Simple disconnection of the cortical venous reflux was not achieved in any of the cases. Three patients with a cavernous sinus fistula were submitted to endovascular treatment and complete occlusion of the fistulas after surgical disconnection of the cortical venous reflux due to persistent ocular symptoms.

There were four complications from the 93 endovascular procedures: one patient had worsening hemiparesis and homonymous hemianopsia due to venous congestion after purposeful partial occlusion of the superior sagittal sinus, one patient had transient increase in the intraocular pressure after embolization of a cavernous sinus fistula, improving in 24 h, with no clinical consequences, another patient had an intracerebral hemorrhage after the first embolization procedure, with minimal residual sequelae and one patient submitted to a diagnostic angiogram died shortly after that, from medical complications unrelated to the procedure.

Surgical disconnection of the cortical venous reflux was the definitive treatment for all the 23 patients. A single surgical procedure was required in 21 patients, and two patients required a second surgical procedure after a postoperative angiogram showed persistence of cortical venous reflux despite intraoperative impression of a successful disconnection. Surgical complications occurred in three patients: a left temporo-occipital venous infarct and hemorrhage resulting in short term memory disturbances, and two related to massive bleeding during the surgical approach. One was a patient who had her fistulae partially resected at the time of the evacuation of an intracranial hematoma. During re-operation for the treatment of the dural arteriovenous fistula a

few years later, significant bleeding from the craniotomy occurred. The same patient had a cerebrospinal fluid leak and a wound infection, requiring further surgical debridement. She eventually recovered well, but was left with persistent homonymous hemianopsia, related to her initial bleeding. The other case of massive bleeding occurred during an attempted disconnection of a complex superior sagittal sinus fistula. Due to the unique architecture of the fistula, massive bleeding was predictable. The patient is alive but severely disabled (seizures and right hemiplegia). Twenty-two patients were well at the last follow-up visit, all with improvement or completely stabilize symptoms (ocular, tinnitus) and no intracranial hemorrhage.

Discussion

Dural arteriovenous fistulas are uncommon lesions. It is estimated that they comprise about 10%–15% of all intracranial arteriovenous malformations. Thought to be acquired, these lesions consist of one or more anomalous arteriovenous connections located in the dura-mater, usually close or within the walls of a cranial venous sinus. More commonly diagnosed in the adult population, they may manifest with a variety of symptoms, including benign ones such as headache, bruit, ocular symptoms (exophthalmos, congestion, and ophthalmoplegia) or potentially devastating ones, such as intracranial hemorrhage and progressive cognitive symptoms as a result of venous congestion [13, 15, 20].

We adopted the Borden et al. [1] classification, where the presence of cortical venous reflux and the relationship of the fistula with the venous sinus are used to determine the category of the fistula. Since the presence of cortical venous reflux (leptomeningeal veins draining the fistula) is accepted as a sign of aggressive behavior (hemorrhage, progressive neurological deficit due to venous congestion), the Borden classification allows easy differentiation between aggressive and benign dural arteriovenous fistulas, guiding well treatment decisions. The validity of the Borden classification for clinical presentation was tested in a large series by Davies *et al.* [5].

The natural history of these lesions started to be clarified in the last decade, and the aggressive behavior of dural arteriovenous fistulas with cortical venous reflux was confirmed in many publications [6, 15, 17, 18, 20–22]. The management of these lesions is still a matter of debate. The treatment of type III lesions by disconnection of the cortical venous reflux was suggested to be safe and effective. It's also recognized that type I lesions

do not have a significant risk of intracerebral hemorrhage and could be managed conservatively, with treatment offered for cases of unbearable local symptoms (ocular symptoms, tinnitus, etc) [16, 20]. However, the idea of converting an aggressive type II (with cortical venous reflux) in a benign type I (without cortical venous reflux) is not accepted by all as an effective treatment, the main reason being that the fistula itself is not completely occluded or resected at the time of treatment, and this may represent a risk for future recanalization and intracerebral hemorrhage.

Disconnection of the cortical venous reflux was suggested by some authors, initially to treat dural arteriovenous fistulas with direct reflux (Borden type III), with the assumption that they should have the same behavior as similar lesions found in the spine [4, 3, 18]. The ligation of the draining vein was appropriately thought to be safe, since the brain was no longer using that vein as a drainage conduit, and the occlusion of this vein would result in complete occlusion of the fistula itself. The management of fistulae where the venous drainage combines cortical venous reflux and sinus involvement (Borden type II) is more complex. The total occlusion/resection of the fistula and the involved sinus will certainly remove the fistulous connections located in the dura, but the resection or packing of a venous sinus involved in the drainage of normal brain tissue may result in catastrophic complications (venous infarct, venous hypertension with cognitive symptoms) and frequently involves complex surgical procedures and significant blood loss [7, 8, 10, 19]. This unique condition

where increased risk of hemorrhage from abnormally “recruited” cortical veins is associated with an impairment of the venous drainage of the brain demands a thoughtful treatment process in order to protect patients from both hemorrhage and venous congestion.

The conversion of a malignant type II dural arteriovenous fistulas in a benign type I is intuitively attractive. Mironov [14] first reported two cases where endovascular techniques were used to perform this kind of treatment. In spite of this initial report, endovascular disconnection is precluded in the majority of the cases because of absence of good endovascular access to the feeders, the sinus and/or the cortical venous drainage. Using endovascular techniques we achieved complete occlusion of the fistula in 12 from 53 patients with type II fistulas (22.6%). None of the simple disconnections were possible using only endovascular techniques. A team approach to these lesions is highly recommended [12]. Surgical exposure of the sinus or even of intracranial veins was used in some of these cases to allow the interventional radiologists adequate access to the fistulas.

The Toronto Brain Vascular Malformation Study Group has adopted the simple disconnection of the cortical venous reflux (Figs. 1 and 2) as the first choice in the management of the Borden type II dural arteriovenous fistulas. Total occlusion of the fistula, if possible by endovascular means with reasonable risk is an option, but extensive open surgical or endovascular procedures in order to occlude or resect completely the Borden type II fistulas in the dura-mater are almost never performed here nowadays.

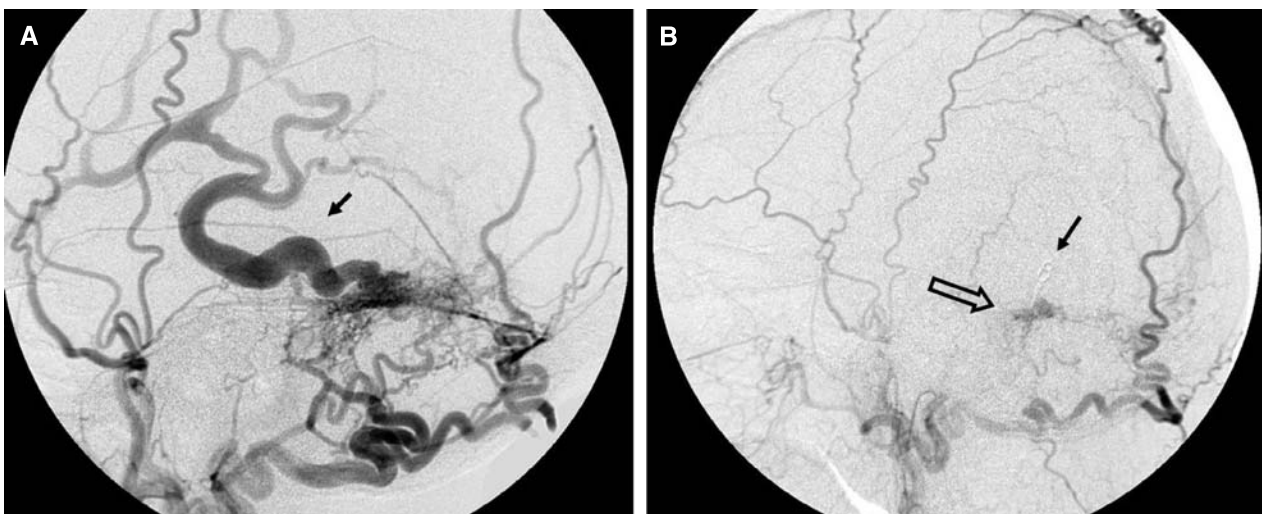


Fig. 1. Pre (A) and postoperative (B) angiogram showing the significant cortical venous drainage (*arrow*) and the remaining fistula (*open arrow*) still present in the dura of the skull base after successful disconnection of the cortical venous reflux. The surgical clip (*arrow*) used to occlude the vein can be seen

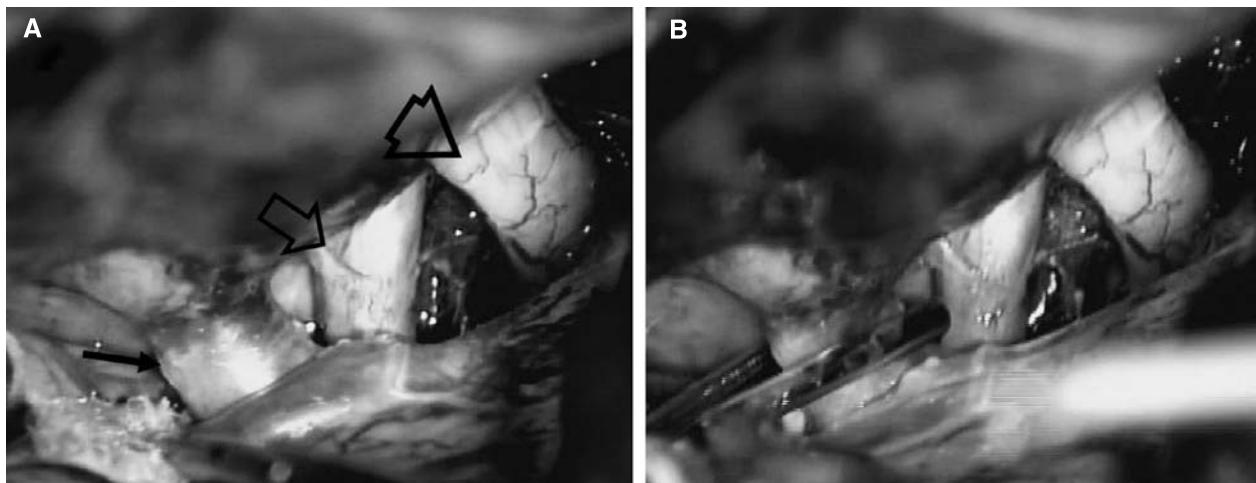


Fig. 2. Arterialized vein draining from a fistula in the region of the left cavernous sinus. Vein (*arrow*), internal carotid artery (*open arrow*) and optic nerve (*arrow head*). Pre (A) and during (B) clipping, illustrating the simplicity of the surgical disconnection

Twenty-three patients submitted to disconnection alone were followed by an average of 4.89 years (2 months to 11 years). At the last follow up, 22 patients were clinically well, with no evidence of intracerebral hemorrhage or recurrent cortical venous reflux. Local symptoms were controlled and/or tolerable in all. Considering the grim natural history of these lesions, with an annual mortality of 10.4% and an event rate (hemorrhagic and non-hemorrhagic neurological deficit) of 15%/year, it is clear that treatment should be offered to all patients. We believe that an average follow-up time of 4.89 years (112.6 patient-years) with no further hemorrhagic event or neurological deterioration after disconnection of the cortical venous reflux suggests that this may be an acceptable treatment for type II dural arteriovenous fistulas, probably as effective as the total occlusion or resection, although associated with fewer complications.

The surgical procedure was associated with permanent complications in only two cases (8% complication rate) including one visual field defect that was most likely due to the hemorrhage at presentation. There was no mortality. A single surgical procedure was enough to occlude the cortical venous reflux in all but 2 patients. The ability to occlude the cortical venous reflux in the first attempt is in our experience related to the degree of understanding of the three-dimensional anatomy of the lesion and its relationship with the skull base and venous sinus. A complete angiogram with spin views and high quality MRI/MRA are very helpful in planning the surgical approach.

The question of recanalization is an issue that we are aware of and all patients with type II dural arteriovenous fistulas that were submitted to disconnection are fol-

lowed regularly at our clinic. Another consideration is the utility of repeated angiograms during follow up, which would have a low but not negligible complication rate, especially in older patients. Digital subtraction angiography is the gold standard for the diagnostic evaluation and treatment decision in DAVF cases, but the availability of a non-invasive method to follow these patients after treatment, or maybe identify or rule out the presence of the cortical venous reflux in the initial diagnostic work up would represent a significant improvement in management. Our group tested the reliability of ATECO-MRI (Auto-triggered Elliptic Centric Ordered 3D gadolinium-enhanced Magnetic Resonance Angiography) in the diagnosis of intracranial dural arteriovenous fistulas, comparing it with conventional digital subtraction angiogram. The overall sensitivity and specificity of ATECO-MRA for the detection of intracranial dural arteriovenous fistulas and the presence of cortical venous reflux was 95% and 89%, respectively (Kurflan, presented at the 8th congress of the WFITN, Venice 2005).

We were able to perform ATECO-MRA in 9 of the 23 patients submitted to a simple surgical disconnection in a late phase of follow up. Cortical venous reflux was absent in all 9 patients, and in one case the persistence of the fistula (but not cortical venous reflux) was suggested by the test. Assuming that the sensitivity of the technique in detecting the presence of cortical venous reflux is high, and that the presence of cortical venous reflux is the major risk factor for future hemorrhage, this long term imaging follow-up adds to our belief that the surgical disconnection is a safe treatment and that recurrence of the cortical venous reflux is not a frequent occurrence. However, due to our limited understanding of the history

of these lesions after disconnection, we think that these patients should still be followed clinically and a standard angiographic examination is warranted when any change in symptoms, including improvement, occurs.

Conclusion

The surgical/endovascular disconnection of Borden type II dural arteriovenous fistulas is an attractive alternative for the management of these lesions. The shorter and much simpler surgical procedure is obviously an advantage. The preservation of the sinus, which in most of these cases is still being used to drain normal brain tissue, is another. The mentioned risks of recanalization of the cortical venous reflux due to the persistence of the fistula in the dura, which would justify a more aggressive approach, seems to be small. We believe that disconnection of the cortical venous reflux should be considered as a valuable option in the management of Borden type II dural arteriovenous fistulae.

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Comment

The authors report their experience with a simple surgical disconnection of 25 dural arteriovenous fistulae (DAVF) Borden type II (cortical venous reflux with involvement of a venous sinus; annual mortality rate of 10.4%) in which embolization was in a first stage attempted (even repeated: 93 procedures with 4 complications) but not successful. Only 23 patients could be followed-up. These patients had a mean age of 53.9 years. In 17 patients the fistula was located at the cavernous sinus so that ocular symptoms predominated. Tinnitus was the presenting symptom in two patients. The surgical procedure seems to have consisted in the simple disconnection of the cortical venous reflux with a clip. Two surgical procedures were repeated and 3 complications occurred. The surgical procedure reached a stable DAVF disconnection in these 23 cases. Twenty-two patients showed a good clinical status at the last follow-up (mean follow-up time: 4.9 years). This report stresses the stability of a simple surgical disconnection of DAVF Borden type II not amenable to an endovascular treatment. Such a treatment should be evaluated and discussed with the patient before repeating an endovascular procedure.

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