Endovascular Approach and Technique for Treatment of Transverse-Sigmoid Dural Arteriovenous Fistula with Cortical Reflux: The Importance of Venous Sinus Sacrifice

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ABSTRACT:

Introduction: Treatment of dural arteriovenous fistula involving the transverse-sigmoid region with cortical reflux is complex; treatment options may require sacrifice of the fistulous segment of sinus.

Methods: We reviewed cases of endovascular sinus sacrifice for dural fistulae at our institution from 2007-2012. Demographic, decision-making, technical, and outcome data were collected.

Results: Seven patients were identified who underwent endovascular sinus sacrifice for treatment of dural fistula during this 4 year period. Determination of the fistulous sinus segment was based on the pattern of cortical venous drainage. Endovascular access to the sinus was achieved trans-arterial, transvenous, or via open surgery in one case. Complete cure of the target fistula was obtained in all cases. One patient had transient post-procedure headache. There were no hemorrhages, new neurological deficits, or sign of increased intracranial pressure. 6/7 patients had angiographic follow up at least 6 months with no recurrence.

Conclusion: Fistulas of the transverse-sigmoid sinuses with cortical reflux may require sacrifice of the parent sinus for cure. Defining the fistulous segment and occluding this segment deliberately, completely and precisely is essential for cure. Several modalities and approaches can be used to achieve this. For properly selected patients, cure of the lesions can be achieved with this method with low risk of morbidity.
INTRODUCTION:

Management of dural arteriovenous fistula malformation (dAVF) is often challenging due to the complex anatomy of the lesions. It is thought that the pathogenesis of most dAVF likely involves a cycle of sinus thrombosis with recruitment of dural collateral arterioles\textsuperscript{1-3}. The pattern of recanalization of the occluded segment then determines the clinical course. If anterograde flow in the sinus results, the patient may experience pulsatile tinnitus, but generally with a relatively benign clinical course\textsuperscript{4,5}. If retrograde flow in the sinus results (Cognard Type IIa), there is increased risk of cortical venous reflux (Cognard Type IIb or IIa+b/ Borden Type II), which is associated with risk of hemorrhage\textsuperscript{6-8}, and carries an annual mortality risk of 10.4\%\textsuperscript{6}.

Dural fistulas in the transverse-sigmoid region with cortical venous reflux represent a unique subset of dAVF which require complete treatment due to the high risk of hemorrhage\textsuperscript{6} and the potentially complex drainage patterns in the cortical veins and contralateral sinus. We present a series of these cases treated with sinus occlusion, integrating the lessons learned from the previous era of predominantly transvenous treatment, with the current technical advances of Onyx (ethylene vinyl alcohol in DMSO: Ev3-Covidien: Irving, CA). In addition, we focus on the decision-making and technical nuances in order to clarify the optimal approach in the contemporary era, where myriad treatment modalities and techniques are available. We emphasize that 1) the direction of cortical venous drainage (not sinus flow direction) determines whether a sinus segment is functional, and that 2) the occlusion should be performed deliberately, completely, and precisely over the nonfunctional segment to avoid inadvertent occlusion of normal cortical drainage.

METHODS:

The endovascular neurosurgery records for the Department of Neurosurgery at our institution were reviewed for a diagnosis of dAVF, over the period of 2007-2011, using an approved Institutional Review Board protocol. These patients were then reviewed to determine those who underwent endovascular venous sinus sacrifice as part of the treatment. Angiographic data including treatment sessions, treatment modalities, angiographic cure, and results of follow up angiography were recorded. Clinical data including age, presentation, timing of treatment, and clinical outcome were recorded. Individual illustrative cases were identified to highlight examples of the endovascular techniques used as well as factors involved in decision making for sinus sacrifice.

RESULTS

Seven patients were identified who underwent endovascular sinus sacrifice for dural fistula treatment during the 4-year period of the review (see table). Age at
presentation ranged from 46 to 63. One patient presented with hemorrhage, and the remainder presented with headaches or other neurologic symptoms. All fistulae involved the transverse, sigmoid, or jugular bulb, and had cortical venous reflux, though one patient (number 7) had a complex fistula involving the contralateral sinus also. Multiple treatment sessions were required for cure of the target fistula in all but three patients. These three of the patients underwent sinus sacrifice as the initial treatment, while transarterial methods for feeder embolization were attempted initially in the remainder. Methods used for sinus occlusion included transarterial embolization with Onyx through arterial feeders and pushed into the sinus (n=1), a transarterial approach to coiling the venous sinus (n=1), transvenous coiling (n=4), and surgical access for coiling an isolated sinus segment (n=1). The illustrative cases describe the decision-making and technique for each approach.

Clinically, all symptomatic patients improved after treatment. One patient remained unchanged who had very complex bilateral fistulae with continued fistula filling on the contralateral side. The target fistula was angiographically cured in all 7 patients. 6/7 of these patients have undergone follow-up angiography at minimum 6 months (range 6-24m). There was no new hemorrhage, venous infarction, or new neurologic deficit post procedure.

*Illustrative Cases:*

**Case 1: Transarterial intravenous sinus embolization with coils, and liquid embolic agents (Figure 1).**

This 62 year old female presented with a left frontal intraparenchymal hematoma measuring 4 x 3 x 2 cm with mild aphasia and right sided hemiparesis. Angiography revealed a large dural fistula located in the region of the left transverse sinus with massively dilated arterial feeders from the left external carotid artery. In addition, there were large feeders from the right middle meningeal, bilateral meningohypophyseal trunks, and a left posterior cerebral artery branch. There was evidence of steal angiographically from the left hemisphere as well as venous ectasia, dilation, and extensive cortical reflux.

The patient was taken one week after presentation for embolization. Initially, the very large dilated segment of vein entering to the junction of the transverse-sigmoid was catheterized trans-arterially. An attempt was made initially to preserve the sinus, and coiling of the dilated venous segment was initiated at the junction with the sinus. After deploying 36 coils and n-BCA through this segment, it was noted that there were several vessels still filling distally directly into the sinus. Two further large pedicles were then embolized with coils and n-BCA glue. A second embolization session was undertaken 6 days later via a large dilated arterial feeder. Though initial attempts were made to preserve patency of the sinus, there was found to be continued fistula feeding directly into the sinus. At this point, an Echelon microcatheter (manufacturer) was advanced trans-arterially through the fistula into a dilated vein, and Onyx was pushed into the sinus in the region of the two distal coiled tributaries resulting in complete occlusion. Control angiography revealed no further filling via any of the vessels and no cortical reflux.
The patient recovered well from the hemorrhage, has undergone multiple follow-up diagnostic angiograms and has been followed clinically for 24 months with no sign of recurrence.

Case 2: Transvenous sinus embolization with coils (Figure 2).

This 48 year old female presented with pulsatile tinnitus, and was found to have a dural fistula involving the distal transverse sinus with complete occlusion of the sigmoid sinus distal to the fistula. Feeding vessels were primarily from the left external carotid artery with smaller branches from the right external carotid and vertebral arteries. There was evidence of cortical reflux into a large temporo-occipital vein with a direct connection to a large superior convexity vein draining into the superior sagittal sinus. Several other refluxing veins were also noted, however a transition in the medial portion of the transverse sinus was noted where there was normal anterograde cortical drainage. This was thought to represent the transition into the functional segment of sinus.

Sinus occlusion was considered the first line treatment for this fistula, which clearly involved the sinus itself. A diagnostic catheter was positioned in the left external carotid artery. The right internal jugular was then catheterized from the femoral vein and a 6French 80cm Shuttle sheath (Cook Medical Inc., Bloomington IN) was positioned. A Neuron 0.053 (Pneumbra Inc., Alameda CA) guide catheter was then advanced across from the right to the left transverse sinus due to distal sigmoid occlusion.

An Echelon-14 (eV3 neurovascular, Irvine CA) microcatheter was then advanced through the sigmoid sinus and into the dilated temporo-occipital vein distal enough to the point of fistulization that complete disconnection of that vein could be achieved. A total of 48 coils were then deployed in order to tightly pack this segment of sinus as well as each of the origins of the cortical veins. Control arterial angiography showed no further filling of the fistula after embolization with preservation of the normal venous drainage into the medial transverse sinus.

The patient’s tinnitus resolved; she had temporary worsening of headache after the procedure, which resolved within one week. She has remained asymptomatic at seven-month follow-up with no sign of recurrence angiographically.

Case 3: Transvenous sinus embolization with coils of isolated sinus segment using surgical sinus access (Figure 3).

This 62 year old female presented with headache and progressive left pulsatile tinnitus. She was found to have a left sided transverse-sigmoid dural fistula fed primarily from left external carotid artery branches. In addition, feeding from posterior cerebral artery branches was noted. The sinus was found to have near occlusion proximally and distally to the segment of sinus with extensive cortical reflux. The feeding vessels were clearly involving this segment of sinus but were too small to achieve adequate complete transarterial embolization of that segment of sinus.
Initially, attempts were made to access the fistulous segment of sinus first from the left internal jugular and then from the left transverse sinus via the right internal jugular. Despite attempts to probe through the occluded segments, it proved impossible to access this segment. Therefore, direct access to the fistulous segment was gained via a craniotomy, with a sheath directly placed into the sinus. A 5 French guide catheter was then passed into this sheath, and an Echelon microcatheter was advanced into the sinus. The entire segment of isolated sinus was tightly packed with a total of 43 coils. Control angiography revealed no further filling and there was no further retrograde venous drainage.

The patient’s symptoms completely resolved and followup angiography at 8 months showed no filling of the fistula.

DISCUSSION:

Treatment of cranial dAVF depends on the anatomic features of the fistula and the patient’s clinical symptoms. Cognard/Borden Type I fistulae involve only the dural sinus with normal anterograde flow and no cortical venous reflux. Treatment is typically considered for symptom (pulsatile tinnitus) amelioration only. Cognard Type II fistulae directly involve the sinus: type IIa with retrograde sinus flow and normal cortical drainage, type IIb with anterograde sinus flow but with cortical reflux, and type IIa+b with retrograde flow in the sinus with cortical reflux. The Borden and Dindjian systems both classify any fistula draining into the sinus with cortical reflux as Type II. Since cortical reflux is the most important risk factor for subsequent hemorrhage, this is the key group to focus on. The key concept for treatment of dAVF is interruption of the exiting venous channel at the earliest point after fistulization. In general we agree with the overall approach as previously outlined. Fistulas involving the transverse-sigmoid region with cortical reflux, however represent a challenging subset to treat safely and completely and is the focus of this series.

Two major paradigm shifts have characterized the endovascular treatment of dAVF. Initial endovascular treatment included a variety of transarterial methods including injection of PVA, silk suture, or liquid embolic, but were primarily performed prior to surgical intervention and did not result in long lasting cure. The advent of detachable coils and detachable balloons allowed for more precise control of embolization and transvenous routes of access became more standard therapy. These series firmly established the concept that only the fistulous segment of sinus should be sacrificed. While this concept was clearly described, only sporadic series including patients with direct sinus fistulas with cortical reflux were reported, the largest of which was reported by Urtasun who reported a 70% cure rate in 10 such patients. Other smaller series reported cure in 1/4, 1/1, and 4/5 patients.

The second major shift then occurred in the mid 2000s with the availability of Onyx (Ev3 Neurovascular: Irving, CA), which allowed for distal penetration into the fistula without a wedged position, and has largely revolutionized the endovascular treatment of dAVF. The Onyx allows for penetration into multiple
feeding vessels, and if there is a parallel channel of fistula within the sinus, it may be possible to treat with sinus preservation, however if the fistula truly involves the sinus segment itself, the only chance of cure is with occlusion of that segment. Onyx may act more unpredictably when transitioning from the arterial feeders into the fistula and unless the venous anatomy and fistulous segment is carefully planned out, inadvertent functional sinus occlusion may result in sacrifice of normal veins. In addition, it may be more difficult to achieve complete cure of the fistula. Recent series with Onyx support this observation. Natarajan 9 reported cure in only 2/5 direct sinus fistulas with cortical reflux involving the transverse-sigmoid sinus with transarterial Onyx. The series reported by Abud 24 supports the observation that sinus occlusion is usually required for this type of fistula, noting that sinus occlusion was required in 65% (of 20) fistulas directly involving the sinus. The series included 10 patients with transverse-sigmoid fistulas with cortical reflux but the results were not reported separately for these patients. Cognard 25 reported an 80% overall cure rate with trans-arterial Onyx embolization in 30 patients, 10 of whom had direct sinus fistulas with cortical reflux, though this subset was not reported separately. Other small series suggest wide variability ranging from 50%23 to 100%26,27 cure rate in patients with direct sinus fistulas and cortical reflux.

Our overall approach and treatment strategy for dAVF of the lateral sinuses with cortical reflux involves 1) defining the involved or nonfunctional segment of the sinus and 2) ensuring complete, deliberate, and precise embolization of that segment. This approach builds on the experience with transvenous embolization, but with the addition of Onyx to the treatment armamentarium, more complete cure rate can be achieved.

**Defining the fistulous segment:**

Fistulas that directly involve the sinus typically require sinus occlusion for complete cure. That said, there are likely a subset of cases where there is a parallel venous channel within the sinus28 which is the point of fistulization, or the fistula may involve a cortical vein very close to the sinus. These two reasons are likely the reason that a minority of fistulas reportedly involving the sinus have been reported to be cured by transarterial embolization without sinus sacrifice24. In most cases the segment of sinus where there is a fistula must be completely occluded to ensure cure12,15,20. The defining feature of the fistulous segment is the direction of cortical drainage, rather than the direction of flow within the sinus15. The entire segment where there is reflux defines the region that can be safely sacrificed since these veins are not draining via that segment, even if their ultimate route of drainage is not clear angiographically. If there is cortical reflux, the drainage is necessarily occurring via other cortical collateral drainage pathways29,30. Furthermore, it is critical that no cortical veins with anterograde flow are sacrificed. This is why we emphasize the need for precise occlusion, especially in situations where normal cortical draining veins may be near cortical veins with reflux.

This principle is illustrated in case 1 and 2 using different embolization methods. In case 1, the transarterial onyx was allowed to fill the sigmoid sinus in order to completely occlude the fistula, however care was taken to avoid occluding the transverse sinus where a small draining vein was noted. Likewise in case 2,
several large dilated cortical veins with reflux were noted along the transverse sigmoid segment, however, there was one vein draining anterograde into the sigmoid sinus. Care was taken to precisely and completely occlude only the segment with refluxing veins and preserve the anterograde cortical drainage; although ipsilateral sinus flow remained retrograde, with the vein draining to the contralateral jugular, the fistula is cured and the cortical drainage remains anterograde into the sinus.

Angiography remains the gold standard in assessing venous drainage pattern in a specific patient. This requires a complete 6 vessel angiogram (including external carotid artery injections) with delayed venous phase examination. The veins in the region of the fistula must be carefully assessed from each injection to determine their presence and the direction of drainage. For fistulae in the transverse-sigmoid region, both supratentorial and infratentorial drainage must be evaluated. One nuance relating to this pertains to the difficulty which can arise in determining the direction of cortical venous drainage as a result of artifactual changes during power injection. If there is a very high flow fistula, the high pressure of the power injection with diagnostic angiography can transmit contrast through the fistula and retrograde into the venous system. We have found two methods to address this issue. First, the direction of cortical drainage is evaluated only after the power injection is complete, and occasionally a reversal of the initial flow in the venous structures is noted. The second potential technique is with the use of quantitative magnetic resonance angiography (Q-MRA) (NOVA; VasSol Inc., River Forest IL). This technique allows for quantitative measurement of flow in the venous structures as well as the directionality. Though we have only begun to apply this technology to decision making in DAVF, there may be a role in the future for detailed venous function evaluation.

Technical Nuances:

Once the functional and non-functional segment has been identified, a strategy must be developed to completely occlude the non-functional segment while preserving the functional segment including all its cortical draining veins. This may not always be the most direct vascular route to the fistula, since a margin is usually needed within the vein for the first few coils to find purchase. Trans-arterial sinus sacrifice with Onyx can be accomplished, but must be done with the same care that transvenous sacrificed is performed. That is to say that sinus occlusion should be performed deliberately in these patients rather than as an unplanned part of the procedure. Entry of Onyx into the sinus and the region of functional sinus must be carefully monitored. The Onyx within the sinus will have less control and no ability to reposition. This strategy is best suited to situations where there is a long non-functional segment. The position of the most proximal normal draining vein should be carefully understood and one might even consider positioning a second arterial catheter in the vessel that defines the functional segment to ensure that the vein is not being encroached upon.

Venous sacrifice is typically, however, performed with trans-venous coil embolization due to the much higher degree of control. Both groins should be
punctured. We typically position a 6F shuttle sheath (Cook Medical Inc., Bloomington IN) in the internal jugular, with the tip positioned at the jugular bulb. The shuttle sheath is used to provide support and avoid guide buckling into the heart. Through this a guiding catheter (Neuron 0.070” or 0.053”, 105 cm or 115 cm (Penumbra, Alameda CA), or distal access catheter (DAC) 0.057” or 0.070” (Concentric, Mountain View CA)) is advanced into the sinus to provide support for the coiling microcatheter. This is especially important when the torcula needs to be crossed with the microcatheter. The guide catheter beyond the torcular region will provide the support and avoid microcatheter herniation into the superior sagittal sinus. The microcatheter is then advanced well beyond the fistula, preferably into the cortical refluxing vein at the far end of the non-functional segment. The placement of the catheter distal to the fistula allows for a small segment where the coils will need to gain purchase before tight packing can be accomplished. The point where the coiling is started is critical since once the sinus is occluded, there will likely not be a venous access point if there is residual fistula after the treatment. The coiling is then continued until the previously identified first vein which drains into the sinus is reached (the functional segment). If incomplete occlusion of the sinus persists, one may consider supplementing the coil mass with Onyx or n-BCA, taking care to not allow any reflux into the functional segment.

A diagnostic arterial catheter is left in the vessel that best defines the non-functional and functional segment of the sinus. This allows for periodic control angiography to assess the adequacy of sinus occlusion, disconnection of the fistula as well as confirm patency of the antegrade draining veins. In addition, we have found that there are some cases where due to the high flow into a fistula, transvenous roadmaps or angiography may not identify a portion of the fistula/sinus connection due to the high pressure and the arterial road mapping is required.

Special access situations may arise due to the complex nature of dAVF and the putative pathogenic mechanism involving venous sinus occlusion. If the segment cannot be reached directly, a contralateral approach via the torcula may be considered, and may in fact be desirable to allow for a longer segment of non-functional sinus to occlude. (See patient 2) In some patients, a trans-arterial approach into the venous system may be considered if the fistulae are large enough. This is distinguished from trans-arterial feeder occlusion of the sinus as described above in that the microcatheter is positioned through the fistula and into the venous sinus for coiling. (Patient 1) When access to a segment of sinus which is completely isolated (no antegrade or retrograde access can be achieved) is required, surgical access may be considered. (Patient 3) A burrhole or small craniotomy is made over the sinus and an armored sheath is placed into the sinus and secured. In this situation, planning is especially critical to ensure that the sheath is placed in such a way that it will allow for complete embolization of the isolated segment. The craniotomy should be performed in the operating room with image guidance and intra-operative angiographic confirmation of the position of the sheath. Hemostasis around the sheath can be achieved with fibrin glue or temporary packing with hemostatic agents. When the sheath is secure and the wound closed, the patient is brought to the angiography suite for coiling to allow for use of biplanar angiography and control arterial injections. Coiling is then completed using the same techniques
described above. These cases are ideally suited for combined surgical/angiography suites. Houdard previously has described the use of direct surgical access to an isolated sinus segment in 10 patients. The fistulous segment was then occluded with coils, glue, or a combination and the fistula was cured in all cases. 5 patients had long-term angiographic follow-up with stable results.

CONCLUSIONS:

Sinus occlusion is the definitive treatment for dAVF involving the sinus with cortical venous reflux. The direction of the cortical veins define the fistulous segment and any anterograde cortical drainage must be preserved regardless of the direction of flow in the sinus. The sinus occlusion should be planned for and the fistulous segment clearly defined prior to any intervention— transarterial or transvenous. The sinus occlusion should be performed deliberately, completely, and precisely to avoid any residual flow in the segment or any occlusion of normal drainage. Multiple approaches including transarterial, transvenous, or direct surgical as well as many embolic materials including detachable coils, Onyx, and n-BCA may all be required for treatment. Using these principles, a high cure rate with low morbidity can be achieved.

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REFERENCES:


