Dural arteriovenous fistulas of superior sagittal sinus: Case report and review of literature

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A case report and review of the literature of 16 dural arteriovenous fistulas (DAVFs) involving the superior sagittal sinus region are presented. In our case, magnetic resonance angiography detected the DAVF with multiple arterial feeding vessels from both external carotid arteries. The patient was successfully treated endovascularly, with complete occlusion of arterial feeders and a total resolution of symptoms.

KEY WORDS
Angiography, arteriovenous fistulas, embolization, magnetic resonance imaging, magnetic resonance angiography, polyvinyl alcohol.

Dural arteriovenous fistulas (DAVFs) constitute 10%-15% of all intracranial arteriovenous malformations [17]. In our experience (of more than 1800 aneurysms and arteriovenous malformations), they form one fifth of intracranial arteriovenous malformations [8]. DAVFs may be found in any dural structures, but are most commonly found in the region of the transverse, sigmoid, and cavernous sinuses [2,5,8,11,12]. DAVFs in the region of the superior sagittal sinus (SSS) are rare. Their diagnosis and treatment may be problematic because of the midline location, eloquent venous drainage, and multiple and bilateral arterial inputs. Since the SSS drains the majority of the venous outflow of the cerebral hemispheres, surgical resection is tolerated only in the anterior third [12]. Intravascular embolization, eventually combined with open surgery, has emerged as an effective treatment for DAVFs in other locations. We report a case of a DAVF in the superior sagittal sinus treated successfully with endovascular embolization, and review 16 cases previously reported.

Case Report
An obese 46-year-old man with mild arterial hypertension had a sudden onset of vertigo, headache, confusion, right-sided homonymous hemianopia, and right hemiparesis. He had a history of blunt trauma of the occiput while playing volleyball a few years ago. Computed tomography (CT) showed an intracerebral hemorrhage in the left parietooccipital region (Figure 1 A). Contrast-enhanced CT revealed abnormal extracerebral vascular structures near the site of bleeding (Figure 1 B). Left internal carotid angiography was normal. Magnetic resonance imaging (MRI) showed, besides the intracerebral clot, enlarged cortical veins with a flow void, indicative of rapid flow (Figure 2 A). In addition, magnetic resonance angiography (MRA) revealed bilateral extracranial thin and tortuous vessels, interpreted as feeding arteries of a DAVF (Figure 2 B). No feeding vessels from the internal carotid arteries or from the posterior circulation were seen. The subsequent conventional external carotid angiography revealed the DAVF in the posterior part of the SSS, fed bilaterally by branches from the external occipital arteries and middle meningeal arteries (Figure 3 A). Venous drainage was through a dilated cortical vein, which drained into the SSS. A varix in the midregion of the cortical vein was observed and thought to be the site of the hemorrhage (Figure 3 B). The absence of the left transverse sinus was also observed. The patient recovered neurologically 1 week after the hemorrhage.

Two months after the bleeding, the left external occipital artery and middle meningeal artery were catheterized selectively using a Tracker® 18 Uni-
Dural Arteriovenous Fistulas

Control angiography 5 months later showed no signs of malformation (Figure 4). The patient continues to be completely free from symptoms.

**Review of the Literature**

Sixteen cases of DAVFs involving the SSS have been reported previously. Unlike the strong female predominance among patients with DAVFs located in other dural sinuses, especially in the cavernous sinus [17], there is an equal gender distribution (8 men, 8 women) with only three patients less than 40 years of age. Six cases presented with hemorrhage (37%). Four patients had subarachnoid hemorrhage and two had subdural hematoma. Most of these patients suffered from hemorrhage during or after the fifth decade. In addition, a number of ethmoidal and anterior fossa DAVFs have been reported [13,19]. They can have their supply from the eth-
External carotid angiography showing the dural arteriovenous fistula. (A) Arterial phase (anteroposterior view) showing the fistula in the posterior part of the superior sagittal sinus fed by branches from external occipital and middle meningeal arteries. (B) Venous phase (anteroposterior view) demonstrating a dilated cortical vein and a varix in the midregion of the cortical vein.

Control angiography 5 months later showing no signs of the fistula.

supply from branches of the superficial temporal artery, internal maxillary artery, anterior falx artery, ophthalmic artery, and occipital artery (Table 1). Cortical veins to the superior sagittal sinus were reported in 4 out of 16 cases. Venous drainage was reported to occur directly to the SSS in 9 cases (Table 2). Varices were reported in 6 cases. Half of the patients were treated surgically (8 out of 16 cases. Table 3). Embolization was performed in 5 cases, followed by surgery in 1 case. In 2 cases, treatment was either refused or not given.

**DISCUSSION**

DAVFs are generally considered to be acquired lesions and not actual malformations; presumably fistulous connections, which mostly develop in a thrombosed dural sinus. The reason for sinus supply from branches of the superficial temporal artery, internal maxillary artery, anterior falx artery, ophthalmic artery, and occipital artery (Table 1). Cortical veins to the superior sagittal sinus were reported in 4 out of 16 cases. Venous drainage was reported to occur directly to the SSS in 9 cases (Table 2). Varices were reported in 6 cases. Half of the patients were treated surgically (8 out of 16 cases. Table 3). Embolization was performed in 5 cases, followed by surgery in 1 case. In 2 cases, treatment was either refused or not given.

**DISCUSSION**

DAVFs are generally considered to be acquired lesions and not actual malformations; presumably fistulous connections, which mostly develop in a thrombosed dural sinus. The reason for sinus

Radiologic findings showing arterial supply of dural arteriovenous fistulas involving the superior sagittal sinus in 16 patients

<table>
<thead>
<tr>
<th>Arterial Supply</th>
<th>Number of Cases</th>
<th>(Bilateral Supply)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Middle meningeal artery</td>
<td>12</td>
<td>(8)</td>
</tr>
<tr>
<td>Occipital artery</td>
<td>5</td>
<td>(1)</td>
</tr>
<tr>
<td>Superficial temporal artery</td>
<td>4</td>
<td></td>
</tr>
<tr>
<td>Vertebral artery</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Posterior auricular artery</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Anterior falx artery</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Not known</td>
<td>4</td>
<td></td>
</tr>
</tbody>
</table>
Venous drainage of dural arteriovenous fistulas involving the superior sagittal sinus in 16 patients

<table>
<thead>
<tr>
<th>VENOUS DRAINAGE</th>
<th>NUMBER OF CASES</th>
</tr>
</thead>
<tbody>
<tr>
<td>SSS</td>
<td>9</td>
</tr>
<tr>
<td>SSS via bridging vein</td>
<td>4</td>
</tr>
<tr>
<td>Varices</td>
<td>6</td>
</tr>
<tr>
<td>Not known</td>
<td>3</td>
</tr>
</tbody>
</table>

Thrombosis remains unclear [2,5,8–10]. DAVFs in the SSS with multiple feeders are extremely rare; only 16 cases have been reported in the recent literature (Tables 1–3).

The mean age of the patients, including our patient, is 51 years (range, 23–78 years). In contrast to DAVFs at other sites, DAVFs involving the SSS seem to be slightly more common among males (9 out of 17, including our case) [1,2,7,8,10,11,13,16,18]. Trauma may be one of the inciting factors associated with the development of DAVFs in men [4]; however, most are idiopathic. Admittedly, at least a mild head injury can be found in nearly every patient. The most common symptoms in the 16 patients were headache (n = 10), aphasia (n = 3), loss of vision (n = 3), seizures (n = 2), hemiparesis (n = 1), and hemiparesis (n = 1). The DAVFs are frequently associated with dilated cortical veins, and often presented with subarachnoid or intracerebral hemorrhage [4,15]. Over one third of the patients presented with hemorrhage (n = 6). DAVFs at the SSS might have a more serious prognosis, due to the high frequency of hemorrhagic occurrence.

De Marco et al have shown that MRI using spin-echo and gradient-echo sequences is useful in the pretherapeutic planning for patients with DAVFs [3]. In the present case, although the spin-echo images correctly revealed the dilated cortical vein, the actual site of the malformation was not identifiable. MRA correctly identified the malformation in the SSS area with multiple feeding vessels from both external carotid arteries. Thus, in agreement with Schuknecht et al we recommend that MRA should be used in connection with MRI in noninvasive evaluation of suspected DAVFs [20]. However, the diagnosis of DAVF is not excluded by a normal MRI or MRA.

The efficacy of transarterial embolization has been previously clearly established in the treatment of DAVFs of the lateral sinus, of the inferior petrosal sinus, and the cavernous sinus [6–8, 12,14,15,21,22]. Halbach et al have previously reported successful embolization of 5 cases of DAVFs in the SSS region [6]. We have used, in most cases of DAVFs, a combined treatment because of failure of total occlusion or revascular pattern [7,11,14,19,21,22]. In our experience, glue provides permanent closure more often than PVA. However, the glue should be injected directly to the fistula site, as distal as possible. This may not be possible because of difficulties in selective catheterization, and PVA particles show better results. In these cases, follow-up angiography is essential to exclude recanalization. The occlusion of the feeders by preservation of the SSS through open surgery is cumbersome, but should be tried if endovascular surgery fails. Embolization of the SSS can be dangerous, and the risks must be carefully analyzed [23]. The risk of a DAVF is dependent on the pattern of venous drainage. That is, a DAVF with unobstructed antegrade sinus flow carries relatively low risk, whereas a DAVF associated with a downstream sinus obstruction, causing venous hypertension and reflux into the parenchymal veins, is more likely to produce infarction or hemorrhage, as in our case. The venous drainage of DAVFs is either to the cortical veins or to the sinus itself, and the occlusion of these large vascular structures carries a high risk. The other factors such as midline location of the SSS and the multiplicity of the feeders makes transvascular obliteration of the nidus more difficult. The timing of embolization may also be important for total success. Like others, we feel that the malformation should be occluded as soon as possible if the patient has had a recent intracranial hemorrhage or has risk factors related to the malformation [6,11]. In our case, the patient could have been treated earlier, 1 week after the hemorrhagic event when the patient had recovered neurologically.
REFERENCES


COMMENTARY

This is a good article. Its merit lies in reminding us that dural AVFs of the superior sagittal sinus are rare, and are frequently associated with intraparenchymal hemorrhage.

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Kurl and colleagues offer a short discussion of the SSS DAVF, introduced by a classic case presentation: parenchymal hemorrhage associated with obstruction of the fistula's venous outflow, as evidenced by the appearance of the cortical veins on the venous angiographic phase. Several points deserve mention:

(1) While MRI and MRA have become useful in demonstrating the pathology, one must remember that a normal MRI and/or MRA does not exclude the diagnosis of a DAVF.

(2) The risk of this condition to the patient depends on the status of the venous outflow. That is, DAVFs with unobstructed antegrade sinus outflow carry relatively low risk, whereas DAVFs associated with a downstream obstruction causing venous hypertension and reflux into parenchymal veins (as is seen in this case) are more likely to produce venous infarction or hemorrhage. Therefore, the venous phase of the angiogram is crucial.

(3) As the authors point out, endovascular therapy has become an important tool in the treatment of this condition, but surgical obliteration of the fistula site may be required in some circumstances.

It would be dangerous to have embolic material traverse the fistula site and obstruct or occlude...