Management strategies for anterior cranial fossa (ethmoidal) dural arteriovenous fistulas with an emphasis on endovascular treatment

Clinical article

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Object. Dural arteriovenous fistulas (DAVFs) of the anterior cranial fossa are rare lesions that can cause intracranial hemorrhage. Authors of previous reports mostly have described open surgical treatment for this fistula type. The authors’ purpose in the present study was to describe their experience with anterior cranial fossa DAVFs, including their endovascular treatment.

Methods. All patients with anterior cranial fossa DAVFs diagnosed and treated in 3 separate institutions during the last 23 years were retrospectively identified. Clinical charts, imaging studies, and procedural notes were evaluated.

Results. Twenty-four patients (22 males and 2 females), ranging in age from 3 to 77 years, harbored 24 DAVFs in the anterior cranial fossa. Eleven patients were primarily treated with surgical disconnection and 2 with radiosurgery. Eleven patients were treated endovascularly; 7 of these patients (63.6%) were cured. In 4 cases of failed embolization, final disconnection was achieved through surgery. In fact, surgery was effective in disconnecting the fistula in 100% of cases. All endovascular procedures consisted of transarterial injections of diluted glue (N-butyl cyanoacrylate [NBCA]), and there were no complications. Brain edema developed around the venous pouch and confusion was apparent after venous disconnection in 1 surgically treated patient. No patient suffered a hemorrhage during the follow-up period.

Conclusions. Disconnection of an anterior cranial fossa DAVF by using transarterial catheterization through the ophthalmic artery and subsequent injection of NBCA is possible with a reasonable success rate and low risk for complications. In patients with good vascular access this procedure could be the treatment of choice, to be followed by open surgery in cases of embolization failure. (DOI: 10.3171/2008.6.17601)

Key Words • anterior cranial fossa • dural arteriovenous fistula • endovascular embolization • ethmoidal fistula • treatment

Abbreviations used in this paper: AVM = arteriovenous malformation; DAVF = dural arteriovenous fistula; DS = digital subtraction; ECA = external carotid artery; ICA = internal carotid artery; ICH = intracerebral hematoma; NBCA = N-butyl cyanoacrylate; OphA = ophthalmic artery; PVA = polyvinyl alcohol; SSS = superior sagittal sinus.

Based on a limited number of available reports, it seems that the majority of this type of DAVFs are treated with open surgery.16,10,13,7,22 There are very few reports regarding the endovascular treatment of this pathological entity.15,10,22 We present our experience in the management of these lesions including their endovascular treatment.

Methods

Data from 3 institutions were combined. From databases we identified patients with anterior cranial fossa DAVFs diagnosed between 1985 and April 2008. All patient records as well as all cerebral DS angiograms, CT scans, and MR images were reviewed.

Patient follow-up was performed clinically and with imaging studies, preferably DS angiography. Magnetic resonance angiography was regarded as an alternative in
cases in which DS angiography could not be performed. The MR angiography technique involved 2D and 3D time-of-flight contrast-enhanced or time-resolved Gd-enhanced acquisitions.

A treatment was considered to be successful when complete fistula disconnection had been achieved with no residual shunt, as verified on DS or MR angiography.

Results

Twenty-four DAVFs of the anterior cranial fossa were diagnosed in 24 patients and treated at our 3 institutions between 1985 and April 2008. Twenty-two patients were male and 2 were female, with an average age of 57 years (range 3–77 years). Among these patients was a 3-year-old child who harbored multiple adult-type DAVFs, only one of which was located in the anterior cranial fossa; the remaining patients were 28–77 years old (average 59 years).

Clinical Presentation

In a majority of the patients the DAVF was symptomatic at the time of diagnosis (18 [75%] of 24). In 6 patients (25%), however, the lesion was discovered incidentally during planar imaging performed for unrelated reasons.

Eleven patients (45.8%) presented with intracranial hemorrhage. All had ICHs—accompanied by intraventricular hemorrhage in 4 cases, subarachnoid extension of blood in 2 cases, and subdural hematoma in 3 cases. The subdural hematoma was ipsilateral to the ICH in 2 cases and contralateral in 1.

Seven patients (29.2%) presented with symptoms unrelated to acute hemorrhage. Two patients (8.3%) had seizures; 1 of them had a parenchymal scar possibly representing evidence of remote subclinical hemorrhage from the fistula. Three patients (12.5%) were examined for headaches; in 1 patient the headaches were accompanied by nausea, vomiting, and confusion. One patient (4.2%) presented with dysgraphia and another (4.2%) with dementia. This last patient also had another DAVF with pial venous reflux.

Angiographic Characteristics

Arterial Supply. In all patients the ethmoidal branches of the OphAs bilaterally supplied the fistulas. In 5 cases there was only a minor contribution from 1 of the OphAs. The ECAs were injected bilaterally in 21 patients and found to contribute to the fistula in 13 (62%) most commonly via branches of the middle meningeal artery. In 1 patient most of the blood supply to the fistula came from the ECA, because the origin of the OphA supplying the fistula was aberrant, originating from the middle meningeal artery as well as receiving retrograde blood supply from the superficial temporal artery and not from the ICA.

Venous Drainage. All fistulas drained directly into the cortical veins and thus were Type III in the Borden classification. Eighteen (75%) drained exclusively via the frontal cortical veins into the SSS. Two drained into the sylvian veins in addition to the major drainage to the SSS. Two drained primarily through the frontal cortical veins into the sylvian veins and the transverse-sigmoid sinus. One fistula drained posteriorly via an inferior frontal cortical vein into the sylvian veins and the posterior aspect of the SSS. Finally, 1 fistula drained via an inferior frontal cortical vein into the basal vein of Rosenthal to the straight sinus and on the other side through the ophthalmic vein into the superior petrosal sinus.

In 11 (45.8%) of the 24 cases there was a venous pouch on the draining frontal vein on its way to the SSS. Only 6 patients among these 11 cases had presented with intracerebral hemorrhage.

Appearance on Planar Imaging

Pretreatment planar images (CT or MR imaging or both) had been obtained in 17 patients. Intracerebral hematoma was evident in 10 of these patients. Large draining veins were seen in 16 (94.1%) of the 17 cases, with a definite venous pouch in 8. In 4 patients there was brain edema surrounding the venous pouch. One patient had undergone time-resolved contrast-enhanced MR angiography, which actually demonstrated the entire DAVF including the enlarged OphA supplying the fistula.

The enlarged veins, venous pouches, and surrounding edema were not visible on CT or MR imaging when the patients underwent imaging studies several months after treatment. The ICH resolved and in its place there was encephalomalacia and gliosis.

Patients With Multiple Cerebral Vascular Malformations

In 2 patients the anterior cranial fossa DAVF was one of many shunts in a multifocal adult-type DAVF. One of these patients was a 3-year-old child.

In 1 patient there was, in addition to an anterior DAVF, a second DAVF located in the foramen magnum as well as a temporal lobe AVM. Three other patients had a brain AVM as well as a DAVF; 1 patient had an AVM in the basal ganglia, a second had a trigeminal AVM, and the third had a mesencephalic AVM. Among the 4 patients who harbored a coexisting AVM, 2 DAVFs were discovered incidentally during a workup for the AVMs, and 2 were revealed after hemorrhage from the DAVF itself and the AVM was a secondary diagnosis.

Two patients had an arterial aneurysm proximal to the fistula. In 1 patient the aneurysm was located on the OphA; this lesion completely resolved 6 months after treatment of the fistula. In the other patient the aneurysm was located on the anterior communicating artery and was most likely related to an AVM this patient harbored rather than to the fistula.

Treatment Options

All patients were subject to treatment by open surgical disconnection, embolization, or radiosurgery. In the last 10 years patients were offered the endovascular approach as the first treatment option, whereas surgery had been the only treatment previously available. Radiosurgery was performed only when patients refused both endovascular and surgical options.

In 11 patients surgical disconnection was the first treatment. Two patients were treated with radiosurgery. Embolization was attempted in 11 patients and was successful.
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(complete fistula obliteration with no residual shunt) in 7 (63.6%). The 4 cases in which embolization was unsuccessful were eventually cured using open surgery.

Surgery. The most common type of treatment was surgery, resolving 15 of the fistulas. Surgery was effective in 100% of the patients treated with this modality either as the primary therapy (11 patients) or after failed embolization (4 patients).

Operations were performed via a frontal or bifrontal craniotomy. The dura-based feeding arteries of the fistula were identified in the cribriform plate, as were the enlarged cortical draining veins. The fistula was subsequently disconnected on the venous side adjacent to the shunting zone.

Radiosurgery. Gamma Knife treatment was undertaken in 2 of the patients (8.3%). Both patients had refused surgery and endovascular treatment. The target of focused radiation was the nidus of the fistula. In 1 patient 27 Gy of radiation was applied to the 90% isodose line, and in the second patient a target of 2.2 ml was irradiated with 25 Gy to the 50% isodose line.

Endovascular Embolization. All embolization attempts were undertaken within the last 10 years by 1 of 4 experienced interventional neuroradiologists. All procedures consisted of transarterial embolization with selective catheterization through the OphA. The microcatheter was advanced well past the origin of the central retinal artery, and the aim was to inject embolic material into the ethmoidal artery past the fistula and into the first part of the draining vein (Fig. 1). The microcatheter most often used was the Magic 1.2 Fr (Balt). In all of the procedures the embolization material was glue (NBCA, Braun). In 1 patient this glue embolization was supplemented with PVA-particle embolization (150–250 µm, Boston Scientific), and in another patient it was supplemented by embolization with PVA particles (500–750 µm, Boston Scientific) and Onyx 18 liquid embolic agent (ev3).

Prior to embolization, glue was mixed with Lipiodol to prolong the polymerization time and to make the glue visible during fluoroscopy. The glue concentration was 16.6% in 3 cases, 20% in 1 case, 40% in 1 case, and 50% in 5 cases. In the 7 successful endovascular procedures the glue concentrations were 50% in 5 cases and 16.6% in 2. In 3 of the 4 unsuccessful embolizations, the glue concentrations were 16.6, 20, and 40%. The procedure in which glue embolization (20%) was topped off with PVA and Onyx had been performed in 1 of these unsuccessful cases. In the fourth unsuccessful embolization the neuroradiologists were unable to achieve the required positioning of the microcatheter and thus aborted the pro-

Fig. 1. Images obtained in a 55-year-old man with a 1-week history of severe headaches, nausea, vomiting, and confusion. A: Axial unenhanced CT showing an intraparenchymal hematoma in the right frontal lobe. B: Diagnostic right ICA angiogram, lateral view, demonstrating an anterior cranial fossa DAVF. The main supply to this fistula is from the right OphA via the right ethmoidal artery. Venous drainage is into an olfactory vein and via a frontal cortical vein into the SSS (arrows in B). C: Angiogram revealing contribution from the right ECA. Relatively minor supply is also noted from the left ICA (not shown). D: Angiogram, lateral view, obtained after a microcatheter was advanced into the right OphA and selective injections were performed, verifying that the microcatheter tip is located beyond the origin of the central retinal artery. The whole nidus and the proximal part of the draining vein were embolized using 50% NBCA. E: Angiogram, same lateral view as in panel D, demonstrating the radiopaque glue cast formed during embolization (arrow). F: Postembolization right ICA angiogram, lateral projection, confirming complete cure of the fistula. Injection of the left ICA and ECA also showed no residual fistula.
procedure prior to using any glue. In this case the fistula was supplied by an aberrant OphA fed primarily by tortuous branches of the ECA.

Patient Follow-Up

Posttreatment follow-up data were available in 23 patients. Digital subtraction angiography was performed in 20 patients and MR angiography in 3 (2 patients had refused DS angiography and 1 patient had renal insufficiency). There was no evidence of residual fistula on imaging studies in any of the cases followed up. No patient’s condition deteriorated clinically, and there was no indication of hemorrhage.

Imaging studies following open surgery were performed between 1 week and 16 months posttreatment (average 5.3 months). The clinical follow-up ranged from 2 months to 6 years (average 2 years).

One of the patients who underwent radiosurgery was lost to follow-up immediately after treatment. In the other radiosurgically treated patient, clinical examination and DS angiography 24 months after the procedure showed complete occlusion of the fistula.

All patients cured by embolization underwent repeated DS angiography between 1 and 6 months posttreatment (average 2.3 months). The clinical follow-up in these patients ranged from 3 months to 5 years (average 15.8 months).

Treatment Complications

A fluctuating level of consciousness developed postoperatively in 1 of the surgically cured patients. This anomaly was attributed to enlarging edema around the venous pouch due to thrombosis. The patient was treated with steroids, his condition improved after 2 days, and he was discharged to rehabilitation with mild confusion. Follow-up MR images showed slow resolution of the edema within a year together with a residual area of postsurgical gliosis. Two years posttreatment the patient began to experience seizures, which were controlled by anticonvulsion medication.

There were no other treatment-related complications.

Discussion

As previously reported, our population of patients with anterior cranial fossa DAVFs is predominantly male. This finding holds in contrast to the opinion that fistulas located anteriorly in the skull are more common in females. Lasjaunias and colleagues have recently suggested a new embryologically based classification for DAVFs.

They found that fistulas located at the lateral epidural space, which includes the lamina cribrosa ossis ethmoidalis, has a strong male predominance, whereas fistulas located at the ventral epidural space are more common in females. Lesions situated at the dorsal epidural space have no definite sex predominance.

The majority of patients were symptomatic at the time of diagnosis (18 [75%] of 24). Eleven (61.1%) presented with intracranial hemorrhage. Six DAVFs were incidentally discovered as the patients were examined with CT or MR imaging for other indications. Anterior cranial fossa DAVFs often harbor a large venous pouch on the draining cortical vein, which is conspicuous on planar imaging. In this study the presence of a pouch was not significantly associated with a hemorrhagic presentation. When a pouch is not present, enlarged cortical veins located in the frontobasal area can be detected as flow voids on MR images. Large draining veins were retrospectively noted on CT or MR images prior to treatment in all but 2 of our cases (94.2%).

Open surgical disconnection of the fistulas has been very successful. Indeed, most of the patients in this study were eventually cured by surgical disconnection with a 100% success rate. Nonetheless, surgery does carry the risks and the inconvenience inherent to frontal craniotomy.

Radiosurgery has been described as a safe and efficient treatment, however, until the DAVF is occluded there is a risk of hemorrhage. Thus, radiosurgery should not be considered as the therapy of choice for these lesions and was performed only in patients who had refused other treatment methods.

The options for endovascular treatment consist of transarterial or transvenous approaches. Various embolization materials can be used, such as glue (NBCA), particles, or detachable coils. It has been suggested that embolization is technically difficult and too expensive. Moreover, in a very recent publication documenting failed attempts to embolize 7 DAVFs, the authors state that this kind of fistula is rarely amenable to endovascular treatment as the supplying vessels are small and tortuous, precluding catheterization with current technology. Yet, our combined experience shows that this therapy is feasible. We were able to exclude 7 (63.6%) of 11 fistulas by endovascular means, with no postprocedural hemorrhages or other complications.

In all of the cases we used glue (NBCA) as the embolic material. Polyvinyl alcohol particles were used in only 2 cases as a supplement to the glue and not as the sole embolization material. In 1 of these cases a cure was not achieved by endovascular means, and the patient was then treated with surgery. In the other case, an endovascular cure was achieved and shown to be durable on a follow-up angiogram obtained 6 months later. Although it is true that PVA, by itself, does not generally produce a durable cure, it can be used as a supplement to eliminate residual minimal flow through a fistula after glue embolization to induce thrombosis and an eventual cure.

Although we favored the use of glue for embolization, in principle other liquid embolic materials, such as Onyx, would be acceptable as long as the same goal was achieved: to reach past the fistula, into the first part of the draining vein, and disconnect the fistula. In our opinion the use of Onyx might be associated with a higher risk of complications as reflux of the material upstream in the parent vessel is a requirement to allow pushing the Onyx forward. In the case of an anterior cranial fossa DAVF, reflux toward the OphA is obviously not recommended. Glue embolization is done with a diluted mixture injected in a forward direction toward the venous side of the shunt. Therefore, reflux is not part of the technique, nor will there be any reflux toward normal collateral anastomoses in the region as might be the case with Onyx.
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In all of our cases the transarterial approach was used. Although a transvenous approach has been successfully performed by other groups, we believe it to be a more complicated route for treating fistulas in this particular location. One must navigate a microcatheter all the way from the puncture site to the most anterior segment of the SSS to reach the draining cortical vein. The transarterial route is usually shorter with easier access via the OphA into its ethmoidal branches. Of course, the transarterial route is not without risk. When using liquid adhesives through the ICA and OphAs, stroke and blindness are the most feared complications, and reflux of embolization material into the OphA absolutely must be avoided.

We found a report in which the authors describe 3 cases of embolization through the branches of the ECA; in none of these cases was the fistula completely obliterated. In our opinion this approach is not optimal given that the ECA supply is only secondary in most cases.

In all but 1 of our endovascular cases the technical difficulty was not access to the OphA but rather getting the glue to pass the fistula and reach the proximal part of the draining vein. Analysis of our data showed that a 50% glue concentration most commonly resulted in fistula disconnection; however, the choice of glue concentration is a subjective decision made by the treating physician and is tailored specifically for each case. The selected concentration depends on, among other factors, the distance between the tip of the microcatheter and the location of the shunt as well as the rate of flow through the fistula. In 1 of our unsuccessful embolization cases the neuroradiologists failed to achieve proper positioning of the microcatheter in the OphA. In this case the OphA originated aberrantly and received blood supply from extremely tortuous branches of the ECA, making endovascular navigation difficult. Another technical point is that all but 5 of the fistulas in this study received significant bilateral arterial supply from the ethmoidal arteries. This bilateral supply is sometimes an advantage because when embolization of 1 ethmoidal artery fails to disconnect the fistula, one can immediately attempt to embolize from the contralateral artery.

Limitations of the Study

The number of attempted endovascular disconnections in this study is relatively low (11 of 24 cases). Although this study includes our experience with anterior cranial fossa DAVFs in the last 23 years, embolizations for this particular fistula have been practiced in our institutions for only the last 10 years, as advances in microcatheter technology reached a level that enabled distal selective catheterization of the OphAs. In most of the patients treated primarily with surgery, the disease was diagnosed more than 10 years ago when endovascular embolization was not yet considered; in the last 10 years, patients have been offered the endovascular approach as the first treatment option.

A potential weakness of this study is that our patient group comprises cases from 3 different institutions and embolization procedures performed by 4 different neuroradiologists. Because of the infrequency of this disease, it was necessary to gather enough cases to reach practical conclusions. All of the interventional neuroradiologists were trained in the same place and have a similar level of experience, and thus we believe that combining their experiences does not introduce much error.

Conclusions

Disconnection of an anterior cranial fossa DAVF with the transarterial injection of NBCA through the OphA is possible with a reasonable rate of success and a low risk of complications. In patients with good vascular access, this procedure could be the therapy of choice, to be followed by open surgery in cases in which embolization fails to provide a cure.

Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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