Dural arteriovenous fistulas misdiagnosed as intracranial neoplasms: illustrative case

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BACKGROUND Dural arteriovenous fistulas (dAVF) may induce imaging findings attributable to various disease entities including malignant neoplasms. In these cases, diagnosis and adequate treatment are often delayed and patients may be exposed to spurious treatments in addition to the risks inherent to an untreated dAVF with cortical venous drainage.

OBSERVATIONS The authors report a case of a patient referred for surgical treatment of a supratentorial high-grade glioma. Thorough review of imaging data challenged the initial radiological diagnosis and led to proper angiographic workup. As a result, a high-grade dAVF was confirmed and successfully embolized. In addition to this case, we provide an extensive literature review on dAVF initially diagnosed as cerebral neoplasms, including clinical, imaging and follow-up data.

LESSONS The literature provides diagnostic criteria for dAVF on magnetic resonance imaging; however, those criteria may be only partly applicable in many cases. Misdiagnosis of a neoplasm due to dAVF has been reported but remains rare, especially in supratentorial lesions. Digital subtraction angiography should be pursued to rule out an underlying vascular pathology if any doubt. This may prevent unnecessary interventions such as biopsies, pharmacological treatment and a delay in dAVF treatment, given its associated risk of hemorrhage and nonhemorrhagic neurological deficits.

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KEYWORDS cortical venous drainage; dAVF; dural arteriovenous fistula; endovascular; glioma; tumor

Dural arteriovenous fistulas (dAVFs) are rare, with a reported incidence of 0.16–0.51 case per 100,000 individuals per year.1,2 The pattern of venous drainage3,4 predicts the clinical course in dAVF. If cortical venous drainage is present, 30% of patients will suffer hemorrhage, corresponding to an annual risk of 6%. Thirty percent of patients (4% annual risk) will present with nonhemorrhagic neurological deficits (NHNDs).5 Although hemorrhage on imaging is usually unambiguous, primary investigation by computed tomography (CT) or magnetic resonance imaging (MRI) in nonhemorrhagic cases may demonstrate findings initially not attributed to dAVF.6,7 This may lead to incorrect diagnoses and erroneous decisions on further diagnostic and treatment measures.

In the case presented here, first a supratentorial neoplasm was diagnosed and referred for surgical treatment. Fortunately, additional imaging revealed the correct diagnosis and led to successful occlusion of a high-grade dAVF. In addition to this case, we also provide an extensive literature review on dAVF initially diagnosed as cerebral neoplasms, including clinical, imaging and follow-up data.

Illustrative Case

A 69-year-old male patient presented without relevant comorbidities or history of trauma, cranial surgery, or infection. MRI done 13 years earlier was unremarkable. While doing sports, the patient experienced a self-limiting generalized seizure. Thereafter, neuro-examination, laboratory tests, and CT (Fig. 1C) were all unremarkable. Despite receiving anticonvulsants, the patient experienced further seizures and was admitted to a...
neurological unit after 8 days. Anticonvulsants were escalated and a noncontrast MRI was performed (Fig. 1A and B), raising suspicions of a left frontal intraaxial tumor. Contrast-enhanced MRI was added (Fig. 2), showing cortical enhancement and increased vascularity and cerebral blood volume. The initial radiological diagnosis was malignant glioma, and the patient was referred to our neurosurgical department for surgery. Further review raised suspicions of a vascular malformation; six-vessel digital subtraction angiography (DSA) then demonstrated a Borden III/ Cognard III dAVF (Fig. 3) fed from the frontal branch of the right middle meningeal artery (MMA). The feeder traversed the midline draining into a left frontal bridging vein with retrograde filling of the cortical vein and prominent filling of a deep choroidal drainage. Also, antegrade drainage of the cortical vein toward the left sphenoparietal sinus was visible. No other feeders were present. A slight dilatation of the draining veins was apparent without significant venous ectasia. Transarterial embolization was performed via a right-sided transfemoral access with a long 8-Fr sheath and a Neuron MAX 088 guiding catheter (Penumbra Inc.) placed in the proximal right external carotid artery (ECA). A Phenom Plus distal access catheter (Medtronic Inc.) was advanced to the proximal MMA, an Apollo 5-cm embolization microcatheter (Medtronic Inc.) was used to inject PHIL 25% (MicroVention Inc.) to the fistulous point. Complete obliteration was confirmed by DSA after the procedure. After treatment, the patient did not experience any neurological deficits, returned to work and was free from seizures with the established anticonvulsant regimen. A follow-up electroencephalogram was within normal limits. On MRI 3 months after embolization, parenchymal findings had disappeared (Fig. 4). Another DSA study will be performed 6 months after embolization to verify total obliteration of the dAVF.

This manuscript was prepared in accordance with CARE guidelines for case reports.

Discussion
A dAVF initially misdiagnosed as a neoplastic pathology is rare, with the majority of cases accounting for brainstem lesions.8–18

FIG. 1. Axial T2-weighted (A) and diffusion-weighted (B) images showing vasogenic edema within the swollen posterior part of the superior frontal gyrus. No engorged veins are visible within the gyrus or on the cortical surface. Native CT (C) of the corresponding area.

FIG. 2. Axial (left) and coronal (right) gadolinium-enhanced T1-weighted magnetic resonance images. Patchy cortical contrast enhancement is visible on the lateral aspect of the superior frontal gyrus, which is slightly enlarged by vasogenic edema. A singular corkscrew-like dilated leptomeningeal vessel (white arrow) traverses the lesion, traveling from the cortical surface to the lateral wall of the lateral ventricle. No clusters of vessels around the dural sinus or venous ectasias are visible, the fistula itself is not perceptible, and veno-occlusive disease was not found.

FIG. 3. Anteroposterior (A) and lateral (B) views of the early arterial phase of a right ECA angiogram. Asterisk depicts frontal branch of right MMA. White arrows mark the fistula point to a left frontal bridging vein with early retrograde filling. Late arterial phase of the same ECA angiogram (C and D). Black arrowheads show deep venous drainage toward the lateral ventricle wall, and white arrowheads mark antegrade drainage via a cortical vein and the sylvian vein toward the sphenoparietal sinus.
Gestion. In the lesions, demonstrating parenchymal changes attributable to venous con-

sider. In the first case, diagnosis of dAVF was already established and surgical treatment was chosen. A biopsy was taken during surgery, exhibiting swollen endothelial cells and nonspecific microglosis but otherwise normal parenchyma. Another study reports a thalamic biopsy for a suspected neoplasm/lymphoma. The non-neoplastic histology was deemed false-negative, as it showed subacute anoxic damage but otherwise intact thalamic histoarchitecture. Eventually the dAVF was visualized on contrast-enhanced CT by coincidence. In another case report, the biopsy of a cerebellar mass showed non-specific reactive changes and the patient was discharged with long-term steroids. A recurrence of symptoms 6 months later led to the correct diagnosis. Roelz et al.17 described CT and MRI findings in a case involving pons, medulla, and cervical cord with a 9-month history of symptoms and requiring multiple interventions.17 This is also the only case in which neither imaging nor symptoms fully resolved. One might speculate whether full recovery of imaging or symptoms or both may indicate that the underlying pathology has been treated sufficiently. A recent study found a 3% recurrence rate of dAVF initially occluded by endovascular treatment, which should be kept in mind. In our review, 3 cases needed retreatment for incomplete obliteration, 1 was due to persistent imaging findings and 1 was due to recurrence of symptoms, whereas technical difficulties caused multiple treatments in the third.

Based on two cases, Ishihara et al. described that the hyperintense signal due to vasogenic edema would normalize faster on the apparent diffusion coefficient (ADC) map than on T2-weighted images. They found this to be of prognostic value whether a dAVF was fully cured or if a residual shunt or recurrence should be evaluated.

We recommend follow-up with both DSA and MRI. In our case DSA was scheduled after 6 months to rule out incomplete obliteration or recurrence. The MRI was scheduled already after 3 months, demonstrating full resolution of the lesion mimicking malignancy, and any ADC and T2 alterations. This also ruled out the sim chance the patient would have both a dAVF and a malignant glioma.

Two more findings specific to this subset of dAVF are noteworthy. Only 6 cases (32%) drained into a dural sinus (2 cavernous; 2 superior petrosal; 1 superior sagittal; 1 transverse sinus), while all others drained into cortical veins, perimedullary veins or had deep venous drainage. This indicates that dAVF mimicking neoplasms represent potentially aggressive fistulas and require immediate embolization. Previous parenchyma findings have completely disappeared.

Only three cases of diencephalic, two cerebellar and two supratentorial lesions have been reported. Table 1 provides the results of our literature review. We only included cases in which an intracranial neoplasm was the erroneous primary suspicion but an underlying cerebral dAVF was later revealed.

Observations

Venous congestion is the true cause of clinical and imaging findings. Of 19 cases reported, four (21%) underwent biopsy of the suspicious lesions, demonstrating parenchymal changes attributable to venous congestion. In the first case, diagnosis of dAVF was already established and surgical treatment was chosen. A biopsy was taken during surgery, exhibiting swollen endothelial cells and nonspecific microglosis but otherwise normal parenchyma. Another study reports a thalamic biopsy for a suspected neoplasm/lymphoma. The non-neoplastic histology was deemed false-negative, as it showed subacute anoxic damage but otherwise intact thalamic histoarchitecture. Eventually the dAVF was visualized on contrast-enhanced CT by coincidence. In another case report, the biopsy of a cerebellar mass showed non-specific reactive changes and the patient was discharged with long-term steroids. A recurrence of symptoms 6 months later led to the correct diagnosis. Roelz et al. described CT and MRI findings in a case involving pons, medulla, and cervical cord with a 9-month history of symptoms and requiring multiple interventions. This is also the only case in which neither imaging nor symptoms fully resolved. One might speculate whether full recovery of imaging or symptoms or both may indicate that the underlying pathology has been treated sufficiently. A recent study found a 3% recurrence rate of dAVF initially occluded by endovascular treatment, which should be kept in mind. In our review, 3 cases needed retreatment for incomplete obliteration, 1 was due to persistent imaging findings and 1 was due to recurrence of symptoms, whereas technical difficulties caused multiple treatments in the third.

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| Case No. | Authors & Year | Age (yrs) | Sex | dAVF Type/ Fistula Location | Initial Suspected Diagnosis | Anatomical Location of Suspicious Lesion | MRI Contrast Enhancement | Symptom Duration Until 1st dAVF Tx | dAVF Tx Addressing Initially Suspected Lesion | dAVF Complete Obliteration After Final Tx | Suspicious Lesion Fully Resolved After Final Tx | Sxs Fully Resolved at Final FU | Duration of Clinical FU | Suspicious Lesion Improved at Final Imaging FU | Imaging FU for Suspicious Lesion Duration |
|----------|----------------|------------|-----|-----------------------------|-----------------------------|------------------------------------------|------------------------|-------------------------------------|--------------------------------------------|------------------------------------------|--------------------------------------------|--------------------------------|-------------------|--------------------------------------------|-------------------------------------------|
| 1        | Present case   | 69         | M   | Cortical vein               | Glioma                      | Frontal lobe                            | Yes                    | 3 wks                               | No                                          | Endovascular                             | Yes                                        | Yes                                        | Yes                                       | 3 mos                           | Yes                       | Yes                           | 3 mos                      |
| 2        | Goldberg et al, 2016 | 56     | F   | Cortical vein               | Neoplasm, vascular          | Frontal lobe                            | Yes                    | Immediate treatment                  | Yes (biopsy at fistula occlusion)          | Surgery                                  | Yes                                        | Yes                                        | Yes                                       | 14 mos                          | Yes                       | Yes                           | 14 mos                     |
| 3        | Ishihara et al., 2009 | 75     | F   | Cortical vein               | Neoplasm                    | Frontal lobe, temporal lobe             | Yes                    | n/a                                 | No                                          | Endovascular                             | Yes                                        | Yes                                        | Yes                                       | 4 mos                           | No                        | 6 mos                        |
| 4        | Borja et al., 2014 | 51     | M   | Vein of Galen (multiple fistulas) | Neoplasm, viral encephalitis | Thalamus (bilateral)                    | Yes                    | 5 wks                               | No                                          | Endovascular                             | Yes                                        | Yes                                        | Yes                                       | 2 mos                           | n/a                       | n/a                          | n/a                        |
| 5        | Matsumura et al., 2008 | 73     | M   | Vein of Galen               | Glial tumor, metabolic encephalopathy | Thalamus (bilateral)                   | n/a                    | 5 mos                               | No                                          | Endovascular                             | n/a                                        | Yes                                        | n/a                                       | 10 mos                          | n/a                       | n/a                          | n/a                        |
| 6        | Sugue et al., 2009 | 51     | M   | Superior sagittal sinus     | Malignant neoplasm, lymphoma | Thalamus (bilateral)                    | Yes                    | 6 mos                               | Yes (biopsy)                               | Endovascular                             | Yes                                        | Yes                                        | Yes                                       | 4 mos                           | n/a                       | 3 mos                        |
| 7        | Bernard et al., 2018 | 65     | F   | Perimedullary vein          | Infiltrative glioma          | Medulla, cervical spinal cord           | Yes                    | 5 mos                               | No                                          | Surgery                                  | Yes                                        | Yes                                        | Yes                                       | 1 mo                            | n/a                       | n/a                          | n/a                        |
| 8        | Chen et al., 2019 | 66     | M   | Perimedullary vein          | Neoplasm, infectious, inflammatory | Pons, medulla                            | Yes                    | 1 mo                               | No                                          | Endovascular                             | No (small residual shunt)                | Yes                                        | n/a                                       | 3 mos                           | n/a                       | Yes                          | 3 mos                      |
| 9        | Crum & Link, 2004 | 35     | M   | Perimedullary vein          | Neoplasm                    | Medulla                                  | Yes “several wks”       | No                                   | Surgery                                    | Yes                                        | Yes                                        | No                                        | n/a                                       | Yes                                       | Yes                       | 3 mos                      |
| 10       | Duan et al., 2017 | 67     | F   | Superior petrosal sinus     | Neoplasm                    | Pons, cerebellum                         | Yes                    | 1 mo                               | No                                          | Endovascular                             | n/a                                        | Yes                                        | n/a                                       | “several wks”                  | Yes                        | “several wks”                | “several wks” |
| 11       | Iwasaki et al., 2006 | 71     | F   | Cavernous sinus             | Malignant neoplasm           | Pons                                     | Yes                    | 5 mos                               | No                                          | Endovascular                             | (twice) + SRS*                          | Yes                                        | Yes                                       | 3 yrs                          | Yes                       | Yes                          | 3 years                   |
| 12       | Le Guennec et al., 2015 | 36    | M   | Perimedullary vein          | Malignant glioma             | Medulla                                  | Yes                    | 2 mos                               | No                                          | Endovascular                             | Yes                                        | Yes                                        | Yes                                       | 1 yr                            | Yes                       | 6 mos                        |
| 13       | Nambu et al., 2020 | 77     | F   | Cavernous sinus             | Malignant neoplasm           | Pons                                     | Yes                    | 2 mos                               | No                                          | Endovascular                             | Yes                                        | Yes                                        | n/a                                       | 5 mos                           | Yes                       | 5 mos                        |
| 14       | Patsalides et al., 2010 | 53     | M   | Yes                    | No                                          | Endovascular                             | Yes                    | Yes                               | Yes                                        | Yes                                       | Yes                                       | Yes                                       | Yes                                       | 9 mos                           | Yes                       | No                          | 3 mos                      |

CONTINUED ON PAGE 5
**TABLE 1. Overview of cases and their characteristics**

| Case No. | Authors & Year | Age (yrs) | Sex | dAVF Type / Fistula Location | Initial Suspected Location | Neoplasm, encephalitis, demyelination | Anatomical Location of Suspicious Lesion | MRI Contrast Enhancement | Symptom Duration Until 1st dAVF Tx | Tx Addressing | dAVF Complete Obliteration After Final Tx | dAVF Fully Resolved After Final FU | Sxs Fully Resolved at Final FU FU Duration of Clinical FU for Suspicious Lesion | Suspicious Lesion Fully Resolved at Final Imaging FU Duration of Imaging FU for Suspicious Lesion |
|----------|----------------|-----------|-----|------------------------------|---------------------------|----------------------------------------|----------------------------------------|--------------------------|------------------------------------|----------------|-----------------------------------------|----------------------------------|---------------------------------------------------|------------------------------------------------------------|
| 15       | Probst et al., 199416 | 40        | F   | Superior petrosal sinus     | Neoplasm                  | Pons, medulla, cervical spinal cord | Pons, cerebellum, thalamus            | Yes                      | n/a                                | No                         | Endovascular + surgery                     | Yes                               | Yes                                               | Yes                                             | n/a                                               |
| 16       | Roelz et al., 201517 | 76        | M   | Perimedullary vein          | Glioma, lymphoma, inflammatory | Pons, medulla, cervical spinal cord | Pons, cerebellum, thalamus            | Yes                      | 9 mos                              | Yes (biopsy)   | Endovascular (twice) + surgery*          | Yes*                              | Yes                                               | No                                               | 3 mos                                             |
| 17       | Weigele et al., 200218 | 53        | M   | Vein of Galen               | Brainstem glioma          | Pons, meancephalon, thalamus (bilateral) | n/a                                    | “several mos”            | No                         | Endovascular                              | Yes                                | Yes                                               | Yes                                             | 6 mos                                              |
| 18       | Cho et al., 201619 | 49        | M   | Cortical vein               | Neoplasm                  | Cerebellum                            | Cerebellum                            | Yes                      | 6 mos                              | Yes (biopsy)   | Surgery                                  | n/a                               | Yes                                               | n/a                                               | n/a                                               |
| 19       | Ishihara et al., 200920 | 68        | M   | Cortical vein               | Ischemia, neoplasm        | Cerebellum                            | Cerebellum                            | n/a                      | No                         | Endovascular (twice)*                     | Yes*                              | Yes                                               | n/a                                               | n/a                                               |

FU = follow-up; n/a = information not available in the reference; SRS = stereotactic radiosurgery; Sxs = symptoms; Tx = treatment.

* Marks cases that needed repetitive treatment due to incomplete initial occlusion or recurrence.
establishment of correct diagnosis and subsequent treatment. Furthermore, the anatomical location of the supposed neoplasm was not indicative of the fistula location. Enhancing brainstem lesions may be caused by dAVF draining into perimedullary veins, cavernous sinus, vein of Galen and transverse or superior petrosal sinus. Two of three bithalamic lesions had dAVF draining into the vein of Galen, while the latter drained to the distal superior sagittal sinus. This report is limited by variable terminology used in some case reports, imaging data limited to singular pictures and follow-up data not provided. To the best of our knowledge, this is the most extensive literature review on this topic, reporting fistula types, course of treatment, symptoms, and radiological findings.

Lessons

The literature provides diagnostic criteria for dAVF in MRI, however, those may only be partly applicable in many cases. Misdiagnosis of a neoplasm due to dAVF has been reported, but remains rare, especially in supratentorial lesions. DSA should be pursued to establish a correct diagnosis and subsequent treatment. Furthermore, the anatomical location of the supposed neoplasm was not indicative of the fistula location. Enhancing brainstem lesions may be caused by dAVF draining into perimedullary veins, cavernous sinus, vein of Galen and transverse or superior petrosal sinus. Two of three bithalamic lesions had dAVF draining into the vein of Galen, while the latter drained to the distal superior sagittal sinus. This report is limited by variable terminology used in some case reports, imaging data limited to singular pictures and follow-up data not provided. To the best of our knowledge, this is the most extensive literature review on this topic, reporting fistula types, course of treatment, symptoms, and radiological findings.

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