JOURNAL OF NEUROSURGERY:
Case Lessons

J Neurosurg Case Lessons 3(19): CASE2243, 2022
DOI: 10.3171/CASE2243

Anterior cranial fossa osseous arteriovenous fistula of the crista galli with bone erosion: patient series

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BACKGROUND Although an anterior cranial fossa dural arteriovenous fistula (ACFdAVF) is thought to have a fistula on the dura near the olfactory groove, the detailed angioarchitecture remains unreported.

OBSERVATIONS In case 1, a 65-year-old man was found to have an asymptomatic ACFAVF. His computed tomography angiography (CTA)-maximum intensity projection (MIP) showed the shunt point in the crista galli (CG), with the intradural drainer penetrating the destroyed bone of the CG. In case 2, a 78-year-old man had a past history of intracerebral hemorrhage and was found to have an ACFAVF. The rotational angiography (RA)-MIP showed the intraosseous fistula in the CG with the drainer passing through a tiny bone defect of the CG. In case 3, a 35-year-old man was investigated for epilepsy. The RA-MIP showed an osseous arteriovenous fistula (AVF) in the anterior cranial base, with the drainer penetrating the skull osteolytic site. In case 4, a 73-year-old woman was found to have an asymptomatic ACFAVF. Her RA-MIP showed the osseous AVF with the drainer penetrating the CG with bone erosion.

LESSSONS All patients were diagnosed with anterior cranial fossa osseous AVF rather than dAVF, with bone erosion in the CG. These findings should be noted at the time of diagnosis and treatment.

https://thejns.org/doi/abs/10.3171/CASE2243

KEYWORDS anterior cranial fossa; osseous arteriovenous fistula; crista galli; bone erosion

An anterior cranial fossa dural arteriovenous fistula (ACFdAVF) is thought to be rare, accounting for approximately 2% of all intracranial and spinal dural arteriovenous fistulas (AVFs) according to a domestic, multicenter survey.[1] The vast majority of ACFdAVFs occur in the dura adjacent to the olfactory groove, arising mainly from ethmoidal arteries with retrograde cortical venous reflux. Since they often cause intracranial hemorrhage or aggressive symptoms because of the coexisting retrograde venous drainage, open surgery has been recommended because of the high cure rate. However, the development of recent technology of high-quality imaging and interventional devices with liquid material or platinum coils has also contributed to less invasive treatment with safety and efficacy.

To date, however, no study of the detailed angioarchitecture including the bony structure associated with the shunt has been reported. The purpose of this study was to analyze neuroradiological imaging findings, such as routine cerebral angiography, maximum intensity projection (MIP) images of rotational angiography (RA), plain computed tomography (CT), and computed tomography angiography (CTA), and to identify the shunt point precisely as well.

Study Description

Of the 65 patients with intracranial and extracranial dAVFs managed at our institution between April 2009 and June 2021, 4 (6.2%) initially diagnosed with ACFdAVFs were investigated. They consisted of 3 men and 1 woman, ranging in age from 35 to 78 years (mean age, 62.8 years). Their initial symptoms were double vision, epilepsy, and vertigo, respectively, in 3 patients; 1 of them had an intracerebral hemorrhage near the lesion 4 years earlier. The remaining 1 patient was

ABBREVIATIONS ACF = anterior cranial fossa; ACFdAVF = anterior cranial fossa dural arteriovenous fistula; AV = arteriovenous: AVF = arteriovenous fistula; CG = crista galli; CT = computed tomography; CTA = computed tomography angiography; MIP = maximum intensity projection; NBCA = n-butyl-cyanoacrylate; RA = rotational angiography; TAE = transarterial embolization; TVE = transvenous embolization; VEGF = vascular endothelial growth factor.

INCLUDE WHEN CITING Published May 9, 2022; DOI: 10.3171/CASE2243.

SUBMITTED January 27, 2022. **ACCEPTED** February 23, 2022.

incidentally found to have the AVF when he developed acute basilar artery occlusion. The radiological diagnosis was based on the findings of magnetic resonance imaging (MRI), CT, MIP of CT, MIP of CTA, and angiography (Allura Xper FD 10/20, Phillips; Infinix VB, Canon Medical Systems Co.). However, 1 patient did not undergo MRI examination due to pacemaker implantation. MIP images from rotational angiography data using an Allura Xper machine were created using a workstation (Interventional Workspot 1.1.0, Philips) and evaluated in detail in 3 of 4 patients. Three qualified neurosurgeons (S.M., H.T., N.E.) participated in reading the RA-MIP images. In particular, this study focused on the MIP images to identify precisely the fistulous point, the angioarchitecture in the restricted area, and the bone condition in the vicinity of the crista galli.

Results

The arteries participating in the fistula included the ethmoidal artery in 4 patients, the sphenopalatine artery in 3 patients, the facial artery in 2 patients, the superficial temporal artery in 2 patients, and the middle meningeal artery in 1 patient (Table 1). The dilatation of the draining vein included the frontal cortical vein in 4 patients and the olfactory vein in 1 patient. The varix of the draining vein was recognized in 2 patients.

It is worthy of special mention that all 4 patients had bone erosion of the crista galli. The single drainage penetrated “the tiny foramen” leading to retrograde venous reflux in all patients. The shunting point was present in the extradural space; in other words, the fistula point was thought to exist in the bone, rather than the dura mater. As for treatment, all patients underwent surgical clipping to obliterate the shunt. All had good clinical outcomes and resumed their previous lives.

Case 1

A 65-year-old man had a past history of laryngeal carcinoma treated by radiation therapy. He was brought to our institution by ambulance because of acute basilar artery occlusion attributed to atrial fibrillation. Endovascular thrombectomy was conducted within five hours from onset, resulting in complete recanalization. Asymptomatic right ACFdAVF arising from bilateral ethmoidal arteries was incidentally discovered at that time (Fig. 1A–C). The AVF drained to the frontal ascending vein accompanying the varix and olfactory vein on the right side (Fig. 1D). MIP of CTA showed the AVF in the crista galli supplied by multiple feeders with the drainage penetrating the bone erosion in the posterior aspect of the crista galli (Fig. 1E and F). Surgical drainer disconnection was performed to obliterate the lesion. Since the angioarchitecture in which multiple small feeders connected to the single draining vein was also recognized in the CG, this lesion was finally diagnosed as an ACF osseous AVF.

Case 2

A 78-year-old man had a past history of intracerebral hemorrhage in the left frontal lobe 4 years earlier and sick sinus syndrome requiring pacemaker implantation a year ago. He visited our institution for the further examination for double vision. The coronal section of his plain CT demonstrated a small bone defect in the left upper side of the crista galli (Fig. 2A). His CTA showed the AVF on top of the crista galli with a venous pouch (Fig. 2B). He underwent cerebral angiography showing the ACFdAVF on the left side supplied by bilateral ethmoidal arteries and the left sphenopalatine artery, with drainage to the frontal cortical vein with a venous pouch of 7 mm (Fig. 2C and D). The RA-MIP horizontal section of the left common carotid injection showed that the fine feeding arteries were closely aggregated inside the crista galli, with a single draining vein penetrating the bone defect to reflux to the intradural space (Fig. 2E–K). Surgical drainer ligation was successfully conducted to eliminate the fistula. This case was also finally diagnosed as an ACF osseous AVF.

Case 3

A 35-year-old man developed an epileptic seizure and visited our institution. His MRI showed an abnormal intracranial vascular lesion. Plain CT showed a bone defect in the transitional area of the crista galli and cribriform plate (Fig. 3A). Angiography demonstrated an AVF in the anterior cranial base arising from bilateral ethmoidal arteries, sphenopalatine arteries, facial arteries, the left middle meningeal artery, and the right superficial temporal artery, with retrograde intracranial venous drainage (Fig. 3B and C). Consecutive images of RA-MIP confirmed the feeding arteries running in the anterior skull base with a single drainer passing through the bone defect, leading to the frontal cortical vein (Fig. 3D–I). The AVF was also diagnosed as an osseous AVF in the vicinity of the crista galli. Surgical drainer ligation was performed to disconnect the shunt.

Case 4

A 73-year-old woman developed vertigo and visited a nearby hospital. Since her MRI showed an AVF in the anterior cranial

TABLE 1. Summary of our four patients with anterior cranial fossa osseous AVF

Case No.	Age (yrs)/ Sex	Rt/Lt	Initial Sx	Feeding Artery	Shunting Point	Bone Erosion	Draining Vein	Classification	Tx
1	65/M	Rt	None	Bilat EA	CG	Yes	Cortical vein w/ dilatation	Osseous AVF	Surgery
2	78/M	Lt	Bleeding diplopia	Bilat EA, lt SPA	CG	Yes	cortical vein w/ dilatation	Osseous AVF	Surgery
3	35/M	Rt	Epilepsy	Bilat EA, bilat FA bilat SPA, rt STA, lt MMA	CG (bottom)	Yes	Cortical vein	Osseous AVF	Surgery
4	73/F	Lt	Vertigo	Bilat EA, bilat STA, bilat SPA, bilat FA	CG	Yes	Cortical vein w/ dilatation	Osseous AVF	Surgery

bilat = bilatateral; EA = ethmoidal artery; F = female; FA = facial artery; M = male; MMA = middle meningeal artery; SPA = sphenopalatine artery; STA = superficial temporal artery; Surgery = craniotomy and drainer clipping; Sx = symptom; Tx = treatment.

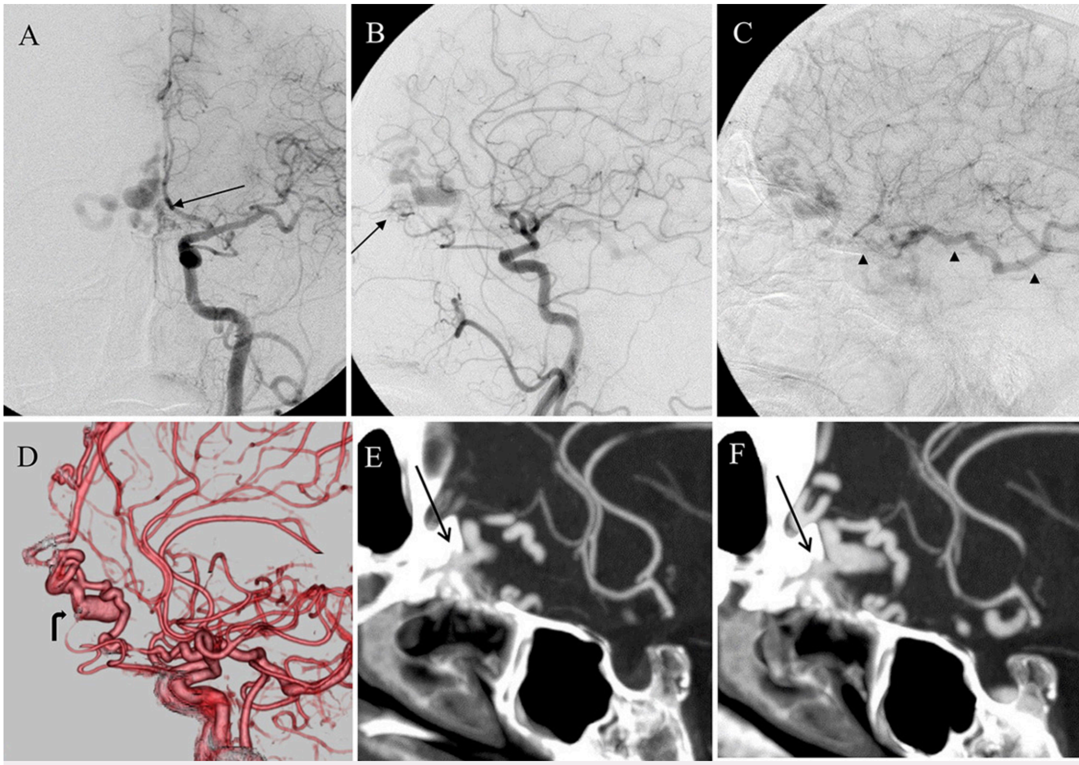

FIG. 1. Case 1. A 65-year-old man with an asymptomatic ACF osseous AVF. Frontal (**A**) and lateral (**B and C**) views of right carotid artery injection showing an AVF (*black arrows*) at the medial aspect of the anterior cranial base arising from the right anterior and posterior ethmoidal arteries, with drainage to the right frontal cortical vein and olfactory vein connecting to the basal vein (*black arrowheads*). CTA (**D**) shows the venous pouch (*curved arrow*) in the frontal ascending cortical vein. The sagittal images of CTA-MIP (**E and F**) demonstrate the AVF in the posterior crista galli, partially destroyed in the posterior aspect, supplied by ethmoidal arteries. Note the fistulous point located in the crista galli (*black arrows*).

fossa, she was referred to our institution. CTA showed a draining vein from the crista galli. However, there was bone erosion where the vein occurred (Fig. 4A). Angiography showed the AVF supplied by bilateral ethmoidal arteries, superficial temporal arteries, sphenopalatine arteries, and facial arteries, with retrograde venous drainage to the left frontal lobe (Fig. 4B–D). Serial coronal images of the rotational right carotid angiogram (MIP) clarified the angioarchitecture in which multiple feeding arteries ran in the skull base to form the AV shunt in the crista galli, with the drainage coursing on the left frontal cortex. The drainer certainly penetrated the bone erosion of the crista galli (Fig. 4E–H). The patient underwent craniotomy to clip the draining vein as close as possible to the shunt point. This lesion was also diagnosed as an ACF osseous AVF in the CG.

Discussion

Observations

Osseous AVFs, known as intraosseous dural AVFs, are rarely reported to occur in parietal bone, sphenoidal bone, petrous bone, occipital bone, etc.[2,3] Recently, Hiramatsu et al.[4] documented many patients with cavernous sinus dural AVFs having the fistulous point in the dorsum sellae or clivus. Mizutani et al.[5] have reported that the shunt points of anterior condylar confluence dural AVFs were present in the skull, based on a radiological study of the intraosseous venous structure adjacent to the jugular tubercle. To date, the concept of bone destruction has basically not existed in dural AVF diseases. However, all 4 of the present patients were found to have bone erosion at the exit of the draining vein. Since routine diagnostic angiography uses subtraction images, bone information is usually eliminated. Therefore, the present study retrospectively investigated the RA-MIP images retaining the bone signal or CTA-MIP images in the bone window, which were carefully reviewed by several qualified neurosurgeons. As a result, the present 4 patients were found to have a small amount of bone destruction in the crista galli. To the best of our knowledge, there have been no similar previous reports, except for 1 patient who had bone erosion of the crista galli.[6] The report described the bony erosion possibly caused by the continuous pulsation of the drainer, but it did not mention the detailed angioarchitecture or the shunt point. Therefore, the present study may be the first report describing not only the extradural shunt, but also the drainer penetrating the anterior skull base leading to the cortical vein. Compared with the thickness of the dura, the skull base is significantly thicker. In addition, cancellous bone pinched by two layers of bone cortex provides an extra three-dimensional space where new blood vessels can spread widely and freely. Moreover, since the crista galli resembles a gulf protruding from the anterior cranial base, intravenous thrombus might tend to occur due to the turbulent flow. Furthermore, there may be a greater chance that the skull sandwiched by the scalp and dura is supplied by not only osseous branches of the scalp artery, but also osseous branches of the dural

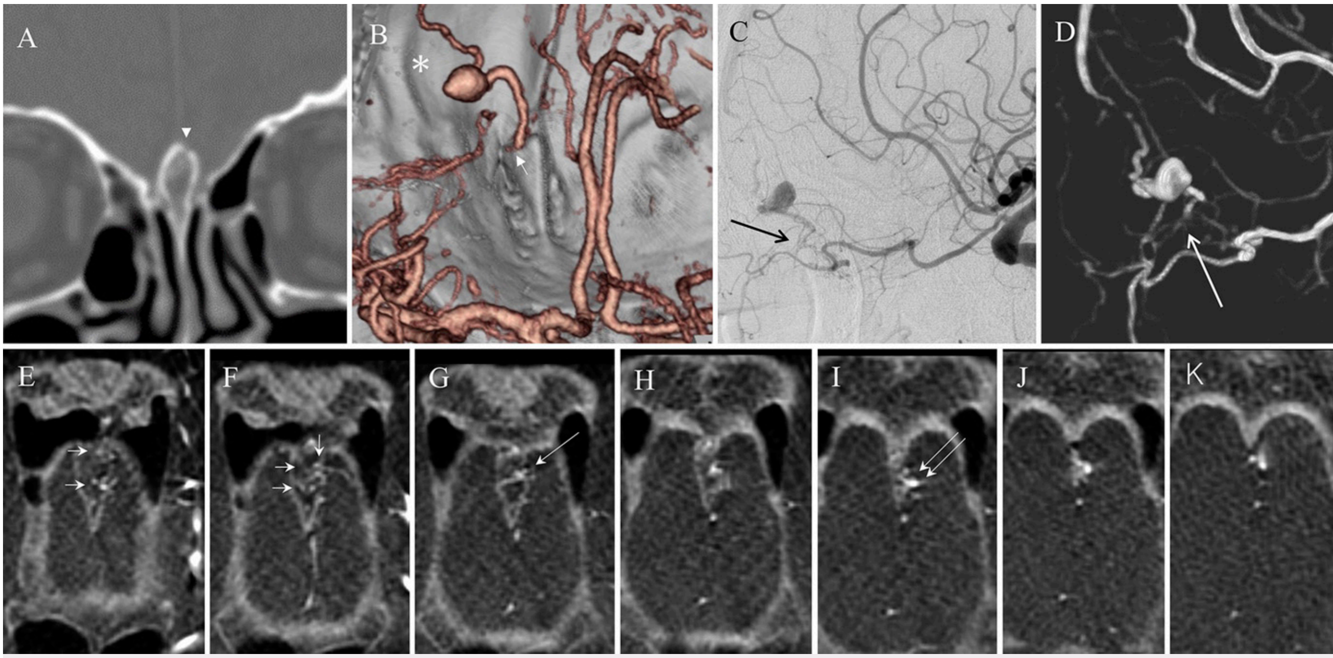

FIG. 2. Case 2. A 78-year-old man with an ACF osseous AVF. **A:** Coronal section of head CT shows the very small bone erosion at the top of the CG (*arrowhead*). **B:** Posterior oblique view of CTA confirms the AVF with the drainer accompanying a venous pouch (*asterisk*), arising from the top of the CG (*arrow*). Lateral view (**C**) and rotational image (**D**) of the left common carotid injection show an AVF in the vicinity of the CG arising from the ethmoidal arteries and right sphenopalatine artery, with drainage to the left frontal cortical vein. The shunt point is indicated by a black arrow (**C**) and a white arrow (**D**). The horizontal sections of RA-MIP with the same injection (**E–K**) show that the tiny feeding arteries coursing in the CG (*short arrows*) participate in the shunt point (**G**, *long white arrow*). The draining vein penetrates the left upper wall of the CG galli (**I**, *long double arrows*).

artery and ophthalmic artery. Thinking of such a situation described above, in fact, we suggest that this area provides satisfactory circumstances for the genesis of an acquired osseous AV fistula.

We believe that there are three points that support the intraosseous shunt theory. First, the vast majority of patients with ACF-dAVFs have an intradural cortical drainage pattern. However, the drainage to the superior ophthalmic vein or nasal bleeding pattern has rarely been documented.[7–9] In other words, the minority of patients have an extracranial drainage pattern. If a shunt point is present inside the dural surface, the draining vein must penetrate both the rigid dura and the skull twice. Since an osseous AVF needs to penetrate only once, it can be easily explained. Second, the sporadic reports of bilateral cortical venous drainage could be explained if the fistula were present in the bone of the crista galli.[10–12] An intradural fistula would not have bilateral drainage because of the presence of the falx cerebri unless two fistulas were to exist simultaneously. Third, other dural AVFs including tentorial dural AVFs or transverse sigmoid sinus dural AVFs are occasionally supplied by pial arteries, but this site is rarely supplied by them, excluding an exceptional case.[13] Thinking of the angioarchitecture of osseous fistulas, it is not easy for pial arteries to penetrate the dura mater and bone cortex to participate in the shunt.

Why Do the AVFs Always Occur in the Midline of the Anterior Cranial Base?

Then, why do these AVFs almost always occur in the midline of the cranial base, not in the lateral skull base? In terms of the etiology of dural AVFs, various factors such as venous hypertension and sinus thrombosis have been considered, and animal experiments support this theory.[14,15] Vascular endothelial growth factor (VEGF) and basic fibroblast growth factor have been proven to play an important role in the genesis of dAVFs based on a histopathological study of surgically resected specimens.[16] However, there are no venous sinuses or developed diploic veins near the crista galli. The present patients had neither a past history of head trauma nor of sinusitis. Olfactory nerves are the only significant structure on both sides of the crista galli.

Neurons and blood vessels are highly associated with each other and generally develop interactively. These two essential systems work based on the transfer of nutrients, oxygen, and information, termed the neurovascular link or neurovascular wiring, and so forth. Although the physiological function at the molecular level remains unknown, several interesting studies have been performed. For example, Mukouyama et al.[17] reported that sensory neurons determine blood vessel branching and arterial differentiation in the skin. Okabe et al.,[18] based on their data using neuronal-specific knockout of VEGFR2 in the retinae of mice, also suggested that neurons control the concentration of VEGF to regulate angiogenesis. Moreover, Li et al.[19] hypothesized a model for nerve-mediated vascular branching and arterial differentiation in developing limb skin. They reported that, after the establishment of a capillary plexus, the pattern of sensory nerves provides Cxcl12 and VEGF-A signals that govern patterns of vascular branching and arterial differentiation during vascular remodeling.[19]

As we know, olfactory nerves are present in the nasal cavity and pass though the ethmoid foramen in the cribriform plate. They penetrate the dura and arachnoid to enter the ventral side of the olfactory bulb.

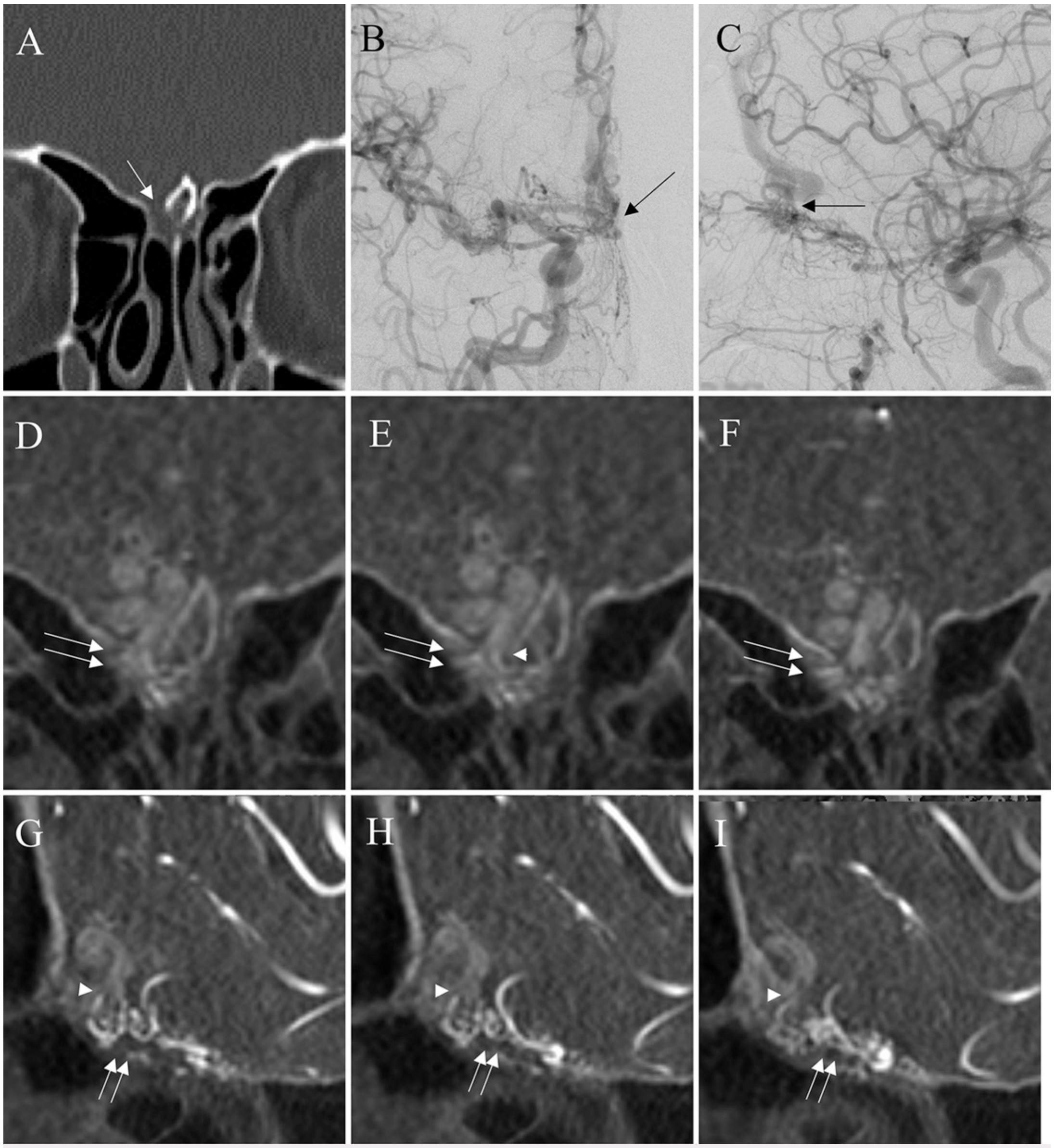

FIG. 3. Case 3. A 35-year-old man with an ACF osseous AVF. Coronal CT (**A**) shows the osteolytic region in the right bottom of the CG (*white arrow*). Right carotid angiogram (**B**, frontal; **C**, lateral) demonstrates the AVF in the area of the midline of the anterior cranial base (*black arrows*) supplied by the ethmoidal arteries, sphenopalatine artery, superficial temporal artery and facial artery, with drainage to the elongated frontal cortical vein on the right side. Consecutive coronal RA-MIP images (**D–F**) delineate the angioarchitecture in which fine arteries (*white double arrows*) feed to form the fistulous point (*white arrowhead*) under the cranial base, leading to the frontal cortical vein. Consecutive sagittal images (**G–I**) display aggregation of small feeding arteries (*white double arrows*) from mainly the ophthalmic arteries with the single ascending drainage that originates from the extradural space (*white arrowhead*).

However, the unilateral olfactory nerve consists of about 20 filum olfactorium, which are thin unmyelinated axons of approximately 0.2 μm in diameter. In other words, as many as 40 delicate nerves are gathered in this narrow part. This number is enough to induce angiogenesis; thus, one could hypothesize that olfactory nerves play an essential role in the genesis of AV fistulas in this nonsinus area. We think a similar mechanism could apply to the genesis of radicular AVFs emerging on the spinal nerve in the craniocervical junction and anterior condylar confluence dAVFs adjacent to the hypoglossal nerve.[20]

Considerations Regarding the Optimal Treatment

To date, surgical drainer ligation has been thought to be the gold standard, with safety and a high cure rate.[21] Recently, several authors have also documented the efficacy of transarterial embolization (TAE) using NBCA (n-butyl-cyanoacrylate) or Onyx.[22,23] However, this method also carries a risk of visual disturbance or visual field defect because a feeder usually arises from the ophthalmic artery. Catheterization to the ophthalmic artery is not always feasible due to severe tortuosity of the vessels. In contrast, recent reports have documented a

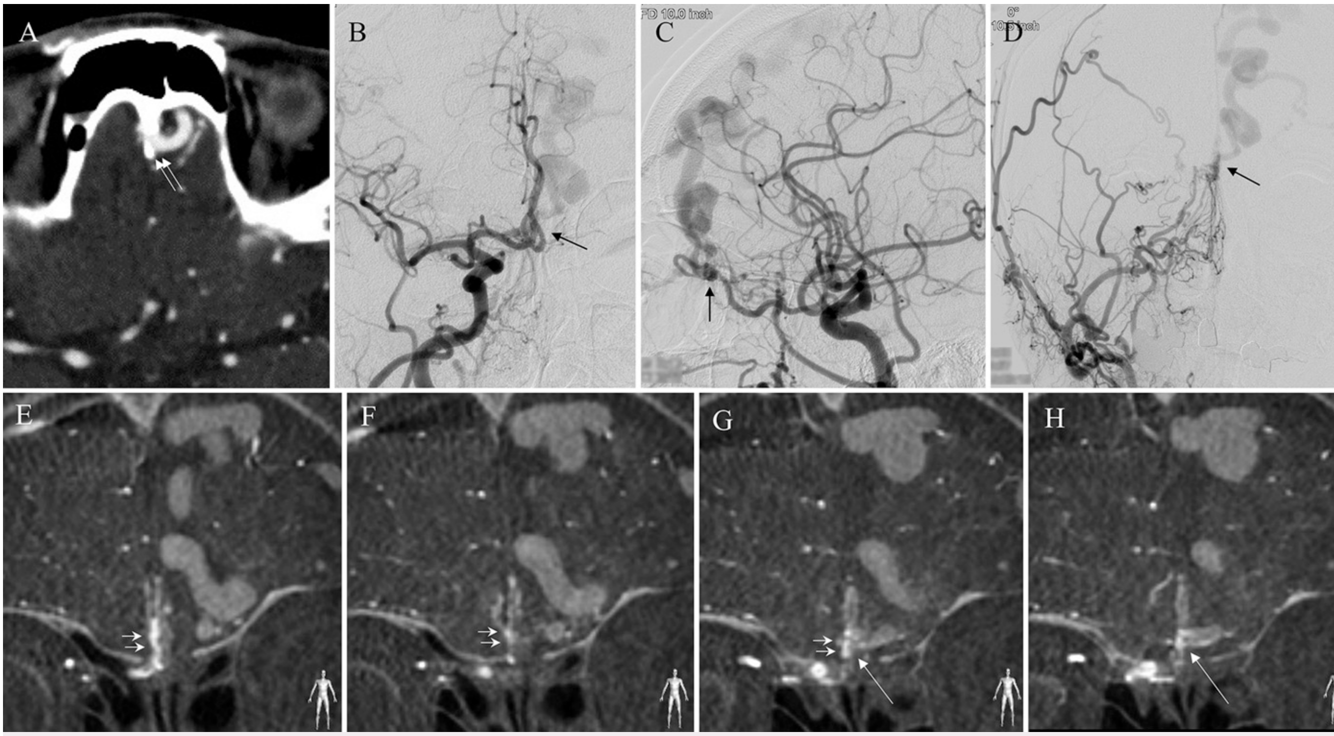

FIG. 4. Case 4. A 73-year-old woman with asymptomatic ACF osseous AVF. CTA-MPR, horizontal section (**A**), shows the single draining vein arising from the eroded CG on the left side (*white double arrows*). Right common carotid angiogram shows an AVF (*black arrows*) near the cribriform plate on the left side (**B**, frontal; **C**, lateral). Caudal view of the selective right external carotid angiogram (**D**) also shows the AVF (*black arrow*) coming from the sphenopalatine artery, the facial artery, and the superficial temporal artery. Consecutive coronal right internal carotid angiography-MIP images (**E–H**) show the AV shunt in the crista galli supplied by multiple feeding arteries (*short double arrows*), with the drainer penetrating the osteolytic region of the crista galli on the left (*white long arrows*). The shunt point is determined based on the obvious contrast change.

higher obliteration rate with transvenous embolization (TVE) using Onyx or platinum coils than TAE.[8,24] If the fistulous point is always located in the osseous region, a transvenous approach could be theoretically curative and safe, because embolization of the intraosseous shunting point using coils or liquid material seems to be reliable and effective. Furthermore, complications related to the ophthalmic artery are predictably less. However, the difficulties in microcatheter control due to the long access length from the puncture site and elongation of the draining vein might carry the risk of venous injury causing intracranial hemorrhage.[25] Therefore, TVE could be the first choice only for selected patients with a good venous access route.

Lessons

The fistula in this disease has been generally understood to be on the dura. However, all of our four patients in the present study, at least, did not have the AVF on the dura mater. Actually, they had epidural osseous lesions in or adjacent to the crista galli accompanying bone erosion. These findings should be noted at the time of diagnosis and treatment. Further investigation and accumulation of this type of data are required in the future.

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Disclosures

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

Author Contributions

Conception and design: Matsubara, Enomoto, Hara, Ogawa. Acquisition of data: Matsubara, Takai, Enomoto, Hirai, Sunada, Yamada, Ogawa. Analysis and interpretation of data: Matsubara, Takai, Enomoto, Tao, Ogawa. Drafting the article: Matsubara, Takai, Ogawa. Critically revising the article: Enomoto, Tao, Ogawa, Uno. Reviewed submitted version of manuscript: Takai, Enomoto, Ogawa, Yagi. Statistical analysis: Ogawa. Administrative/technical/material support: Ogawa. Study supervision: Ogawa, Uno.

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