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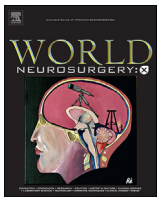

Co-occurrence of dural arteriovenous fistula and meningioma: A rare case and systematic review

Abhishek S. Bhutada [a,b], Srijan Adhikari [b,*], Joshua Cuoco [b], Alexander In [a,b], John Entwistle [b], Mark R. Witcher [b]

[a] Virginia Tech Carilion School of Medicine, Roanoke, VA, USA
[b] Division of Neurosurgery, Department of Surgery, Carilion Clinic, Roanoke, VA, USA

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ABSTRACT

Background: The coexistence of meningioma and dural arteriovenous fistula (dAVF) is a rare, but highly complex condition. Various pathophysiological mechanisms underlie intracranial meningiomas with continuous or distant dAVFs. We describe a case of coexisting meningioma and dAVF with a systematic review of the literature.
Result: Including the present case, there are 21 reported cases of coexisting intracranial dAVF and meningioma. The patients' ages ranged from 23 to 76 years, with a mean age of 61 years. The most common presenting symptom was headache. The dAVFs were commonly located at the transverse-sigmoid sinus (43%) and superior sagittal sinus (24%). The most common meningioma locations were the tentorium and parietal convexity. In 76% of the cases, the meningioma occluded the sinus. The most common dAVF treatment was transcatheter arterial embolization, followed by tumor resection (52%). Among the 20 cases with available outcome data, 90% reported favorable outcomes.
Conclusion: This report highlights some of the features of coexisting dAVF and meningioma and presents a systematic review of other reports on this phenomenon. Through an in-depth analysis of the literature, we highlight some of the leading theories regarding the causes of concomitant dAVF and meningiomas. Our report supports one of the leading theories that impaired venous return, whether through the occlusion of sinuses or sinus manipulation during surgery, plays a role in the development of dAVF. Further understanding may help guide future clinical decision-making and surgical planning.

1. Introduction

Intracranial meningiomas and dural arteriovenous fistulas (dAVFs) are distinct conditions that, in rare cases, can coexist but are seldom reported. Therefore, data on their clinical profile, treatment options, and patient outcomes are limited.

Meningiomas are among the most common types of primary intracranial tumors. Although these tumors are benign in most cases, they can have an insidious clinical course and may persist for years before being diagnosed. These tumors are in the arachnoid layer of the meninges and arise from meningothelial cells. Although the exact etiology is unknown, a few of the reported risk factors include age, female sex, ionizing radiation exposure, and several genetic syndromes.[1,2]

dAVFs are abnormal shunts between intracranial dural arteries and the venous system. Among intracranial arteriovenous pathologies, dAVFs account for approximately 10–15% of cases.[3] The underlying etiology of dAVFs remains unclear. Many factors, including congenital anomalies, have been associated with their occurrence. A few reported cases linked their existence to acquired factors, such as trauma, surgical invasion, infection, or malignancies, leading to the development of venous sinus obstruction and subsequent hypertension.[4]

Here, we describe a patient who was incidentally found to have a coexisting high-flow dAVF while undergoing surgical treatment for left parietal meningioma. Only 20 published case reports or series of this coexistence have been reported in the literature. We provide a detailed literature review highlighting patient demographics, presenting symptoms, clinical features, and treatment courses to better understand this rare occurrence. We also propose some mechanisms that might lead to the pathogenesis of concomitant dAVFs and meningiomas.

* Corresponding author. 1906 Belleview Ave, Roanoke, VA, 24014, USA.
E-mail address: sadhikari@carilionclinic.org (S. Adhikari).

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2. Case presentation

A 40-year-old man presented to the emergency department with a 1.5-week history of transient weakness in the right arm and leg. He had a longstanding history of seizures and pulsatile tinnitus that was never evaluated. MRI brain revealed a large mass in the left parietal brain [Fig. 1]. The lesion caused a moderate mass effect, but there were no signs of midline shift or apparent falcine herniation. Signs of thrombosis in the superior sagittal sinus were also not observed. The patient was started on dexamethasone (2 mg) orally and referred to the neurosurgery clinic for treatment. The treatment options were discussed with the patient and surgical resection of the left parietal mass was planned.

The resection of the left parietal brain mass had to be aborted intraoperatively because of unexpected hypervascularity of the bone overlying the tumor, which compromised the safety of the procedure due to the added risk of hemorrhage. The patient was monitored on an inpatient basis while the interventional radiology team was consulted to perform diagnostic angiography and possible subsequent embolization [Fig. 2]. Angiogram results showed a left parietal meningioma supplied by distal branches of the middle meningeal artery (MMA), anterior cerebral artery, and middle cerebral artery. An incidental finding of high-flow dAVF was also noted. The dAVF was primarily reported to originate from the transcalvarial left occipital branches, but it fed into the posterior aspect of the superior sagittal sinus.

Given the nature of the hypervascularity of the lesion and its proximity to a high-flow dAVF, transcatheter arterial Onyx embolization of the dAVF was performed prior to surgical resection of the meningioma. Embolization of both the MMA and high-flow dAVF was performed. The high-flow dAVF was occluded which resulted in spontaneous regression. The tumor had a large vascular pedicle with a vein draining directly into the sagittal sinus.

Three days post-embolization, the patient underwent gross total resection of the left parietal mass. The patient tolerated the procedure well, without any intraoperative complications. The final pathology report confirmed the diagnosis of meningioma with a World Health Organization classification histologic grade of 1.

Postoperatively, the patient only experienced mild right hemiparesis, which resolved within a few days prior to discharge. His postoperative magnetic resonance imaging (MRI) results showed complete resection of the mass, with no residual enhancing lesions in the area. The patient underwent rehabilitation due to weakness at an inpatient pain medicine and rehabilitation center.

The patient continued to follow-up with outpatient neuroendovascular team that recommended one year angiogram to evaluate recurrence. On repeat angiogram, a complex recurrent dAVF at a similar location in the mid-to posterior-sagittal sinus the following year. Unlike the earlier occurrence, this fistula was supplied by the superficial temporal and middle meningeal arteries [Fig. 3]. There was no evidence of

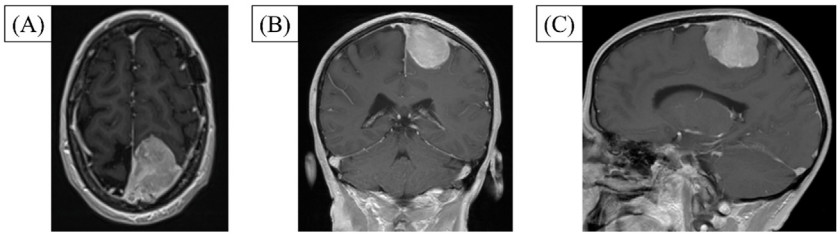

Fig. 1. Pre-operative magnetic resonance image demonstrating a large left parietal avidly-enhancing hypervascular extradural lesion with a dural tail and focal mass effect. A) Axial view. B) Coronal view. C) Sagittal view.

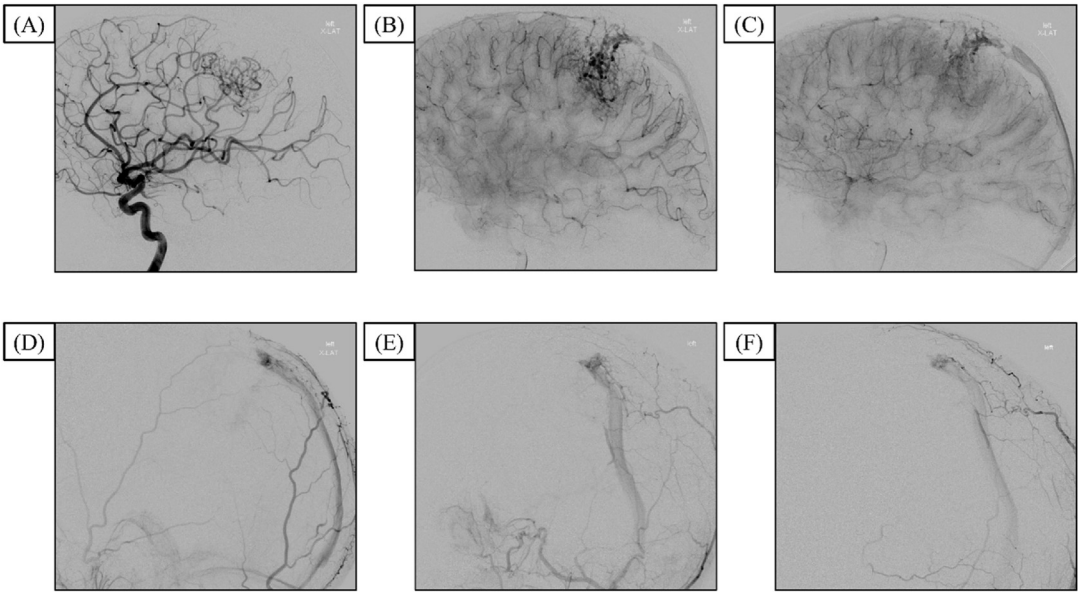

Fig. 2. Initial diagnostic cerebral angiogram demonstrating an incidental dural arteriovenous fistula adjacent to but separate from the hypervascular extradural lesion. A-C) Selective angiography of the left internal carotid artery demonstrates a significant amount of pial supply to the tumor via the distal branches of the anterior and middle cerebral arteries and a large venous pedicle draining the tumor into the posterior portion of the superior sagittal sinus. D-E) Selective angiography of the left external carotid artery demonstrates an enlarged occipital artery with distal transcalvarial branches supplying a dural arteriovenous fistula with early venous drainage into the posterior superior sagittal sinus, adjacent to but separate from the tumor. F) Super selective angiography of the left occipital artery demonstrates distal transcalvarial branches supplying a dural arteriovenous fistula.

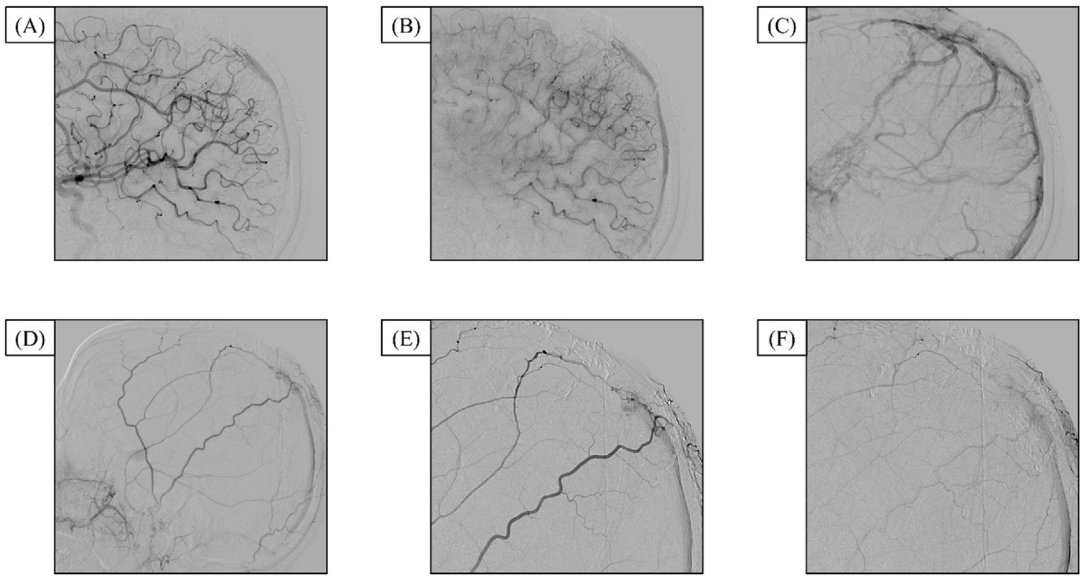

Fig. 3. One year follow-up diagnostic cerebral angiogram. A-C) Selective angiography of the left internal carotid artery demonstrates distal anterior cerebral artery branches extending to form pial collaterals to the dural arteriovenous fistula in the mid/posterior sagittal sinus. D-F) Selective angiography of the left external artery demonstrates an enlarged branch of the superficial temporal artery and middle meningeal artery filling the dural arteriovenous fistula via transosseous collaterals. No evidence of residual or recurrent feeders from the previously embolized occipital artery.

prior feeders from the occipital artery that had been embolized during the last procedure. The patient underwent another transcatheter arterial Onyx embolization of the left MMA. Post-embolization, an approximately 60–75% reduction in flow across the dAVF was observed. No residual shunting was observed in the right external carotid artery branches. However, some residual fistula filling was evident in the branches of the anterior cerebral artery. Overall, the patient tolerated the procedure well. The patient continued to undergo annual diagnostic angiography to monitor the residual fistula filling. At the 3-year follow-up visit, the diagnostic angiogram showed complete resolution of the dAVF, with no evidence of residual fistula filling.

3. Result

A systematic review of the PubMed and SCOPUS databases was conducted using the keywords “dural arteriovenous fistula” and “meningioma.” Articles that were published between the databases’ date of inception until 1/30/2023 were considered. All case reports and series of patients with both dAVFs and meningiomas were collected and analyzed independently by the authors and crosschecked for accuracy. Articles were excluded if they were about any vascular malformation other than dAVF, intracranial tumors other than meningiomas, or dAVFs and meningiomas that did not coexist but occurred as separate temporal events. A total of 118 articles were identified through the database searches, among which 20 articles met the inclusion criteria and were included in the qualitative analysis [Fig. 4]. Each relevant article was thoroughly reviewed to collect information on patient demographics, presenting symptoms, clinical features of each lesion, treatment provided, and outcomes.

A total of 21 cases of coexisting intracranial dAVFs and meningiomas have been reported in the literature, including the present case [Table 1]. The patients’ ages ranged from 23 to 76 years, with a mean age of 61 years. Our review included 11 males and 10 females. Patients most often presented with headaches (52%), tinnitus (24%), and seizures (14%). The dAVFs were commonly located at the transverse-sigmoid sinus (43%) and superior sagittal sinus (24%). The most common locations for the meningiomas were the tentorium (29%), parietal convexity (14%), frontal convexity (14%), and parasagittal convexity (14%). In 76% of the cases, the sinus was occluded by the meningioma.

The surgical treatment strategy was mentioned in each report. The most common treatment was transcatheter arterial embolization of the dAVF, followed by tumor resection (52%). Five patients (24%) underwent tumor resection alone. Three patients (14%) underwent shunt dis connection followed by tumor resection. One patient underwent tumor resection, followed by dAVF resection. One patient was managed conservatively because they were asymptomatic at presentation. Among the 20 patients for whom outcome data were available, 90% had favorable outcomes. In total, two patients had unfavorable outcomes, including the patient in the current case. The other patient who had an unfavorable outcome experienced worsening consciousness and dysphagia immediately postoperatively. In our case, the patient had recurrent dAVF 1-year postoperatively.

4. Discussion

dAVFs are the most common acquired lesions that can be caused by various underlying conditions. The current literature on this topic has identified a rare association between dAVFs and intracranial tumors. The majority of these cases reported that the tumor involved was a meningioma.[5–8] One of the leading theories proposes a causative relationship between meningiomas obstructing the dural sinuses or inducing dural sinus thrombosis and subsequent development of abnormal dAVFs.[9] However, this was not true for all cases of coexisting meningiomas and dAVFs.

There were still a few reports of dAVF formation in patients with no or only partial occlusion of the sinus.[10–12] This was also true in our case, in which MRI showed no signs of sinus occlusion. One theory that might explain the cause of dAVFs in the absence of sinus occlusion is that tumor-related angiogenesis leads to the release of angiogenic factors, such as growth factors and cytokines, that might play a large role in the development of dAVFs.[13] Another leading theory that might explain this phenomenon is that tumors, such as meningiomas, are highly vascularized. Their high vascularity poses a risk for the formation of dAVFs due to arteriovenous shunting through the highly vascular tumor bed.[11,14,15]

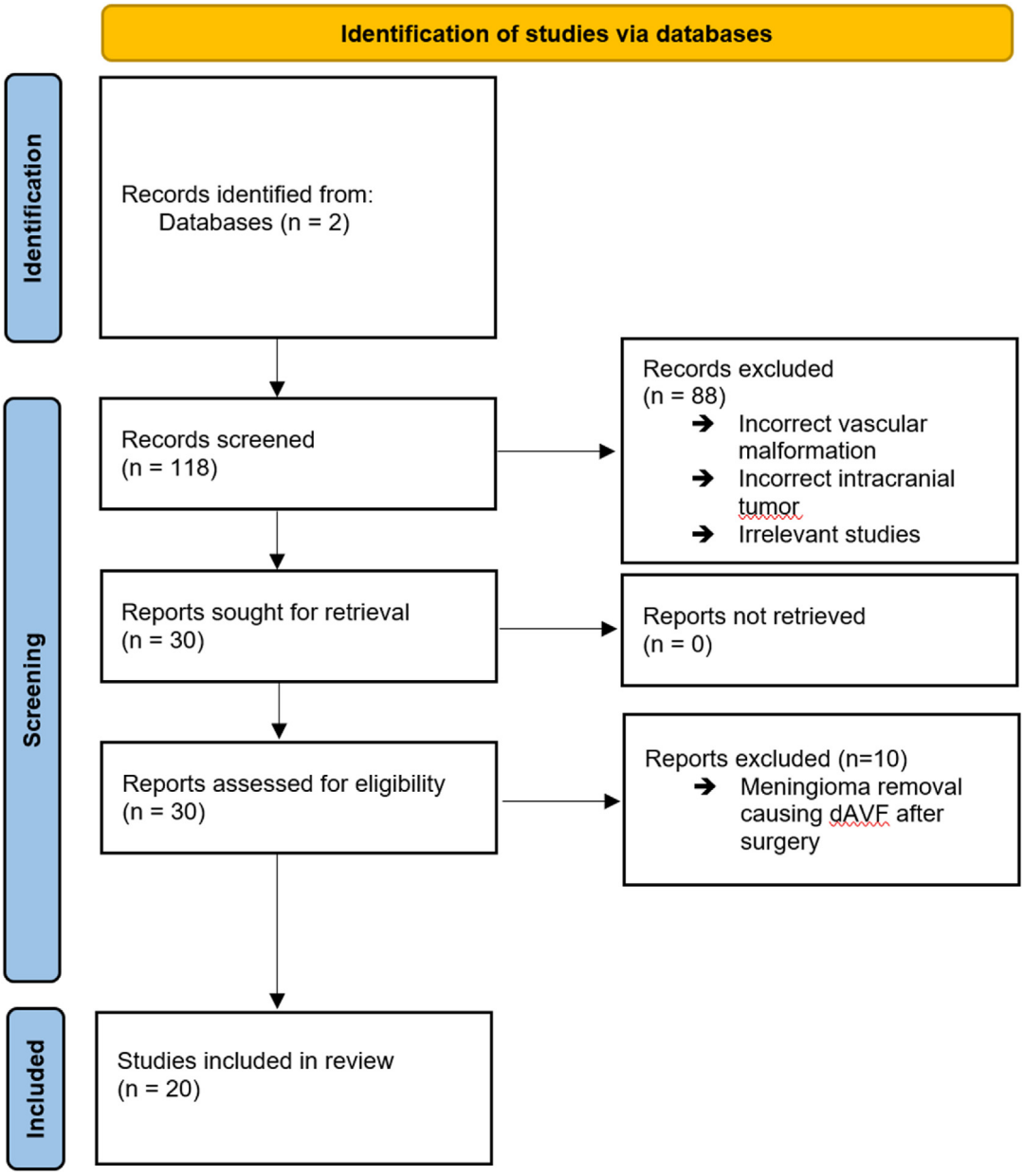

Fig. 4. PRISMA flow diagram for the identification process of the relevant studies.

This might have contributed to the development of dAVF in our patient, since he initially presented with a hypervascular tumor during surgery.

In the present case, it was unusual for dAVF to recur after the completion of transcatheter arterial embolization and complete surgical resection of the meningioma. Recurrence of dAVF has never been reported in any of the cases of concomitant dAVF and meningiomas. A few studies have reported that the resection of a meningioma leads to the development of dAVF.[16–20] In these studies, the meningioma was most often near or invading the dural sinus. These cases demonstrate that sinus manipulation during intracranial surgery carries a potential risk of dAVF development. In our patient, the meningioma was highly vascularized, with a vein draining directly into the posterior aspect of the superior sagittal sinus. This might be one of the reasons that lead to the recurrence of dAVF after meningioma resection. Although dAVF is a rare occurrence, it should still be considered in patients with hypervascular tumors that are near to or invading the dural sinuses that will require sinus manipulation during intracranial surgery.

5. Conclusion

The relationship between intracranial tumors and vascular malformations remains unclear. This case report highlights some of the clinical nuances that might lead to the coexistence of dAVF and intracranial meningiomas. It also supports the leading theories that impaired venous return, whether through the occlusion of sinuses or sinus manipulation during surgery, plays a role in the development of dAVF. Understanding this rare occurrence is critical to guide the diagnosis and treatment of future patients. Neurosurgeons should consider the risk of dAVF in cases that involve sinus manipulation when planning surgical procedures.

Table 1
Summary of case reports describing patients with concomitant dAVF and intracranial meningioma.

Author	Publication Year	Patient Age	Patient Sex	Presenting Symptoms	Side	Site of dural AVF	Site of Meningioma	Sinus Occlusion	Proximity of Lesions	Treatment	Outcome
Preul et al	1992	72	M	asymptomatic	L	Superior Sagittal Sinus	Frontal Convexity	No	Remote	observation	favorable
Yokota et al	1993	65	F	vertigo	R	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Arnautovic et al	1998	63	F	headache, localized pain in left orbit, left paranasal sinus pain, and constant tinnitus	L	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Chung et al	1999	62	F	headaches, tinnitus	R	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	tumor resection	favorable
Vilela et al	2001	51	M	headache, blurred vision	R	Superior Petrosal Sinus + Parietal Convexity	Parasagittal	Yes	Adjacent	shunt disconnection + tumor resection	favorable
Vilela et al	2001	56	M	headache, right hearing deficit	R	Transverse-Sigmoid Sinus	Cerebellomedullary cistern	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Horinaka et al	2003	69	F	headache, tinnitus	R	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	tumor resection	favorable
Ahn et al	2003	45	F	headache	L	Transverse-Sigmoid Sinus	Parietal Convexity	No	Remote	tumor resection	favorable
Inoue et al	2007	76	M	headache	L	Sphenoid ridge	Cavernous Sinus	No	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Toledo et al	2010	60	M	increasing confusion, word-finding difficulties	L	Superior Sagittal Sinus	Parasagittal	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Enatsu et al	2012	76	F	asymptomatic	R	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Copeland et al	2013	60	M	pulsatile tinnitus, headaches, nausea, vomiting	R	Superior Sagittal Sinus	Falx	Yes	Adjacent	tumor resection	favorable
Kusira et al	2014	23	M	headache, papilledema	BL	Sinus Confluence	Sinus Confluence	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Hattori et al	2014	70	M	dysphasia	L	Transverse-Sigmoid Sinus	Sphenoid ridge	Yes	Adjacent	transcatheter arterial embolization + tumor resection	unfavorable
Labidi et al	2015	59	M	headaches, left hand paresthesia, seizures	R	Vein of Trolard	Parietal Convexity	Yes	Adjacent	shunt disconnection + tumor resection	favorable
Quan et al	2019	66	F	headache	L	Transverse-Sigmoid Sinus	Tentorium	Yes	Adjacent	tumor resection	NR
Takemoto et al	2019	59	F	transient aphasia, dysgraphia	L	Anterior Cranial Fossa	Frontal Convexity	Yes	Adjacent	shunt disconnection + tumor resection	favorable
Manzo et al	2020	74	F	tiredness, left-sided hemiparesis	R	Anterior Cranial Fossa	Frontal Convexity	Yes	Adjacent	tumor resection + fistula transection	favorable
Rubio et al	2020	66	F	left eye loss of vision	L	Falcotentorial	Posterior Parafalcine	Yes	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Nakazawa et al	2022	71	M	seizure	R	Superior Sagittal Sinus	Parasagittal	No	Adjacent	transcatheter arterial embolization + tumor resection	favorable
Current Case Report	2022	40	M	seizure, pulsatile tinnitus, weakness	R	Superior Sagittal Sinus	Parietal Convexity	No	Remote	transcatheter arterial embolization + tumor resection	unfavorable

*dAVF, dural arteriovenous fistula.

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CRediT authorship contribution statement

Abhishek S. Bhutada: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **Srijan Adhikari:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **Joshua Cuoco:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **Alexander In:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **John Entwistle:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. **Mark R. Witcher:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Abbreviations list

dAVF	dural arteriovenous fistula
MMA	middle meningeal artery
MRI	magnetic resonance imaging

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