Case Report

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Microsurgical Treatment of Cranial Dural Arteriovenous Fistulas: A Case Series and Literature Review

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ABSTRACT

Dural arteriovenous fistulas are pathological vascular malformations within the dura mater, characterized by abnormal connections between pial arteries and veins. These are rare lesions that can present with a variety of neurological symptoms, and when ruptured, can result in a fatal hemorrhage. This disease can be managed by endovascular embolization, surgical resection, and stereotactic radiosurgery. Particularly with the advancement of endovascular techniques, surgery is less frequently used to treat these lesions. However, it has remained a viable treatment option in specific cases. In this article, we present a series of 17 patients with dural arteriovenous fistulas who were treated in a single tertiary neurosurgical center. We also present a comprehensive literature review regarding the various treatment options for this complex pathology.

Introduction

Dural arteriovenous fistulas are pathological vascular malformations situated in the dura mater, characterized by abnormal connections between pial arteries and veins [1]. Although dural arteriovenous fistulas are relatively infrequent within the spectrum of neurosurgical disorders, they can manifest with a wide range of symptoms, from mild neurological deficits to severe ones, including potentially fatal hemorrhages [2]. Historically, neurosurgical resection has been the primary treatment approach for these lesions; however, contemporary treatment paradigms now encompass various modalities, including endovascular embolization and stereotactic radiosurgery. Advances in endovascular techniques have led to embolization becoming the first-line treatment option for dural arteriovenous fistulas. Nevertheless, microsurgical resection remains an important component of the treatment strategy, particularly in select cases where it may be considered the optimal approach [1,2]. (Table 1) In this article, we present a case series involving 17 patients diagnosed with dural arteriovenous fistulas who were treated at a tertiary neurosurgical center in Portugal. Additionally, we provide a comprehensive review of the current literature concerning the available treatment options for this condition.

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Table 1: Global population social, demographic, clinical, and radiological characteristics.

	Sex	Age at the time of diagnosis	Radiological Presentation	Borden/ Cognard	Arterial feeders	Main venous drainage	Associated vascular malformations	Pre-operative embolization	Clinical Presentation	Neurological presentation	Follow-up time (days)	Clinical outcome	Post-op complications	Radiological outcome
1	M	61	Non-hemorrhagic	III/IV	MMA and STA	Transverse sinus	-	Not possible	Symptomatic	No focal neurological deficits. Headche	431	No neurological deficits	-	Complete exclusion
2	M	63	Hemorrhagic	III/V	Vertebral artery	SSS	PICA aneurysm	Partial	Symptomatic	No neurological deficits, headache	3511	No neurological deficits	Hydrocephalus	Complete exclusion
3	M	85	Hemorrhagic	III/IV	Ethmoidal artery	SSS	Venous aneurysm	Not performed	Symptomatic	Dysarthria	629	No neurological deficits	-	Complete exclusion
4	M	77	Hemorrhagic	III/III	MHT	Petrous sinus	AICA aneurysm	Not performed	Symptomatic	Central facial palsy, dysarthria, dysmetria	55	Death	-	Complete exclusion
5	M	63	Non-hemorrhagic	III/III	ACA	SSS	Multiple aneurysm	Not performed	Symptomatic	No neurological deficits, headache	949	Anosmia	-	Complete exclusion
6	M	63	Hemorrhagic	III/IV	OA and MMA	Transverse sinus	-	Not performed	Asymptomatic	No neurological deficits	571	Epilepsy	-	Complete exclusion
7	M	77	Non-hemorrhagic	III/III	OA and MMA	SSS	Venous aneurysm	Not possible	Asymptomatic	No neurological deficits	326	Right hemiparesis	-	Complete exclusion
8	M	44	Non-hemorrhagic	III/III	Ethmoidal artery	SSS	-	Not performed	Symptomatic	No focal neurological deficits, headache	857	No neurological deficits	-	Complete exclusion
9	M	66	Hemorrhagic	III/IV	Pericallosal artery	SSS	Venous aneurysm	Not performed	Symptomatic	Coma (GCS 6)	1380	Aphasia, PFP, hemiparesis	-	Complete exclusion
10	M	51	Non-hemorrhagic	III/IV	MMA, SCA, AICA	Petrous sinus	Giant venous aneurysm	Partial	Symptomatic	PFC and Loss of sensation	1117	Partial hearing loss	-	Complete exclusion
11	M	49	Hemorrhagic	III/IV	MMA	SSS	Venous aneurysm	Not performed	Symptomatic	Left Hemiparesis	1443	Hemiparesis, Epilepsy	Deep surgical site infection	Complete exclusion

12	M	57	Non-he-mor-rhagic	III/IV	MMA	AVM	Com-plex AVM	Partial	Sympto-matic	No focal neurological deficits, headache	592	No neuro-logical defi-cits	Super-ficial surgi-cal site infec-tion	Late recons-titution of fistula sites
13	M	37	He-mor-rhagic	III/IV	MHT	Cere-bellar cortical vein	-	Not perfor-med	Sympto-matic	No focal neurological deficits, headache	302	No neuro-logical defi-cits	-	Com-plete exclu-sion
14	F	65	He-mor-rhagic	III/V	OA	Bul-bome-dullary vein	-	Not perfor-med	Sympto-matic	Gait instabi-lity and left hemiparesis	242	Gait insta-bility	-	Com-plete exclu-sion
15	M	46	Non-he-mor-rhagic	III/V	Ver-tebral artery	Bul-bome-dullary vein	-	Not perfor-med	Sympto-matic	Paraparesis	195	Para-pare-sis	-	Com-plete exclu-sion
16	M	45	Non-he-mor-rhagic	III/IV	OA, STA and MMA	SSS	-	Not perfor-med	Sympto-matic	No focal neurological deficits, headace	228	Foot Loss of sen-sation	New onset sensory deficit	Parcial exclu-sion
17	M	60	Non-he-mor-rhagic	III/IV	Etmoi-dal artery	SSS	-	Not perfor-med	Sympto-matic	No focal neurological deficits, headache	24	No neuro-logical defi-cits	-	Com-plete exclu-sion

Methods

This study was conducted at São João University Hospital in Porto, Portugal. We retrospectively collected data from all patients who underwent microsurgical or endovascular treatment for dural arteriovenous fistulas between 2014 and 2023.

Inclusion Criteria

1. Patients older than 18 years of age.
2. Patients with confirmed dural arteriovenous fistula on angiography.
3. Patients who have been treated with surgical resection of the fistula or endovascular embolization.
4. Patients whose treatment modality has been selected by a multidisciplinary meeting between neurosurgeons and interventional radiologists.

Exclusion Criteria

1. Patients younger than 18 years of age.
2. Patients who chose not to pursue either open surgical or treatment with embolization.
3. Patients whose lesion was confirmed not to be a dural arteriovenous fistula on angiography imaging.

In total 17 patients have been included in the study. All patients included had been discussed in a multidisciplinary team meeting, consisting of vascular neurosurgeons and interventional neuroradiologists, where the specific features of the case were considered an indication for this specific therapeutic modality. If the open neurosurgical treatment was chosen, the patient underwent a craniotomy and a resection of the dural arteriovenous fistula. If the endovascular embolization was chosen, the patient underwent an endovascular embolization of the fistula via trans arterial or transvenous route. In the case of failure to treat the lesion by embolization the surgical treatment was then pursued. The dural arteriovenous fistulae at the time of diagnosis, according to the angiographic imaging, were classified according to the Borden and Cognard scales classifications, shown in Tables 2 & 3.

- The data collected: Social, demographic, clinical, and radiological data.
- Surgical outcome: post-operative neurological status, post-operative complications, and radiological results. Patient cohort demographics, clinical presentation, and radiological characteristics. The whole cohort is displayed in Table 1 and summarized in Table 4. The patient outcomes were then grouped into primary and secondary outcome groups. The primary outcomes were measured as successful or unsuccessful exclusion of the dural arteriovenous fistula and the resolution of the pre-procedure neurological deficits. The

secondary outcomes were measured as complications such as late-onset hydrocephalus, postoperative wound infections, and new-onset neurological deficits.

Table 2: The Borden classification [3].

Type	Description
Type I	Direct drainage into a dural sinus without cortical venous reflux
Type II	Direct drainage into a dural venous sinus with a retrograde flow into cortical veins
Type III	Direct drainage into cortical veins

Table 3: The Cognard classification [4].

Type	Description
Type I	Antegrade drainage into a sinus or meningeal vein
Type IIa	Retrograde drainage into a sinus or meningeal vein
Type IIb	Reflux into cortical veins
Type II a+b	Reflux into both sinus and cortical veins
Type III	Direct cortical venous drainage without venous ectasia
Type IV	Direct cortical venous drainage with venous ectasia
Type V	Spinal venous drainage

Table 4: Summary of demographic and radiological characteristics.

Sex n (%)	M	16(94.1)
	F	1(5.9)
Median age (years)		61 (47.5 – 65.5)
Radiological presentation n (%)	Hemorrhagic	9(52.9)
	Non-hemorrhagic	8(47.1)
Clinical presentation n (%)	Symptomatic	15(88.2)
	Asymptomatic	2(11.8)
Borden Classification n (%)	III	17(100)
Cognard Classification n (%)	III	4(23.5)
	IV	10(58.8)
	V	3(17.7)

Patient characteristics

A comprehensive overview of patient characteristics is detailed in Tables 1 & 4. The cohort predominantly comprised male patients, accounting for 94.1% of the population. The median age at the time of treatment was 61 years. A significant portion of the cohort presented with a hemorrhage on the initial CT scan, with 52.9% experiencing hemorrhage compared to 47.1% that did not. Focal neurological deficits were observed in the majority of patients, constituting 88.2% of the cohort. All dural arteriovenous fistulas in this study were classified as Borden Class III (100%). Utilizing the Cognard classification system, the most frequently observed lesion type was classified as Cognard IV (58%), followed by Cognard III, which was identified in 23.5% of cases (n=4), and Cognard V, observed in 17.7% (n=3).

Results

Primary Outcomes

Endovascular treatment had been previously attempted in 5 patients (29.4%), resulting in partial embolization of the dural arteriovenous fistulas in 3 patients (17.6%). In 2 patients (11.8%), embolization was attempted but deemed technically unfeasible. The remaining cohort, comprising 12 patients (70.6%), underwent surgical intervention as the primary treatment modality. The decision to pursue surgical treatment was influenced by factors such as the inability to treat the lesion endovascularly and the specific anatomical location of the dural arteriovenous fistula. Additionally, the presence of an intraparenchymal hematoma necessitating evacuation also influenced the need for hematoma evacuation and microsurgical treatment of the AVF. The reasons why endovascular embolization was not completed are summarized in Table 5. The median follow-up in this cohort was an average of 666 days. After surgical treatment was completed, a follow-up DSA was performed during the same hospital stay. Immediate postoperative angiography showed that 94.1% (n = 16) of patients achieved complete exclusion of the lesion; one patient presented with signs of persistent shunting on the postoperative DSA. A second surgery was considered as the treatment option, but the patient refused surgical treatment, and observation with serial imaging over time was pursued instead.

Table 5: Dural arteriovenous fistulas treated primarily through microsurgery. 12 patients in total.

Reason	N (%)
Multidisciplinary decision – inability to endovascular treatment	5(29.4)
Other indications for surgery (ex.: need of evacuation of intracranial hematoma)	5(29.4)
Dural arteriovenous fistula location (located in anterior cranial fossa)	2(11.7)

At the time of the latest follow-up, a second DSA was performed, and 88.23% (n = 15) of patients had shown complete obliteration of the lesion. In one patient, late reconstitution of the fistula sites was observed. For this patient, adjuvant endovascular embolization was chosen as the treatment modality. In the case of resolution of pre-procedure neurological deficits, 41,17 % of patients (n=7) had resolution of their pre-operative symptoms. 58,8 % (n=10) have remained symptomatic after the surgical procedure. One patient has died after the procedure due to a massive pulmonary embolism in the postoperative period (5,8 % n=1).

Secondary Outcomes

The cumulative surgical complication rate in this cohort (Table 4) was 23.6% (n = 4). Complications encountered included surgical site infections (11.8%, n = 2), one case requiring revision of the surgical site. Late-onset hydrocephalus was encountered in one patient

(5.9%), and a ventriculoperitoneal shunt was implanted. New-onset neurological symptoms were observed in a single patient who developed mild paresthesia in one foot, which improved over time. The postoperative complications are summarized in Table 6.

Table 6: Postoperative complications. 4 patients in total.

Description	N (%)
Surgical site infections	2(11.8)
Late hydrocephalus	1(5.9)
New neurological-deficit	1(5.9)

Illustrative cases

Case I

An 85-year-old male presented to the emergency department complaining of a sudden onset of headache. Upon examination, she was alert, oriented, and GCS of 15. No focal neurological deficits were observed during the neurological examination. A head CT was performed, and a right frontal intracerebral hematoma protruding into the subdural space was discovered. Due to the unusual location of the ICH, CT angiogram and later DSA were performed for differential diagnosis. A right frontobasal dural arteriovenous fistula was confirmed, receiving feeders from an ethmoidal artery and associated with a giant frontobasal aneurysm. The venous drainage was observed to be through the superior longitudinal sinus (Figure 1A). The patient was discussed in a multidisciplinary team meeting, and the surgical treatment option was chosen due to the location in the anterior cranial fossa, ethmoidal type of the dural arteriovenous fistula, and the frontobasal aneurysm that it was associated with. The patient underwent a right unilateral frontal craniotomy and exclusion of the AFV. A DSA was performed after surgery and showed a complete exclusion of the dural AVM. (Figure IB). Postoperatively, no focal neurological symptoms were observed. The post-operative course was uneventful.

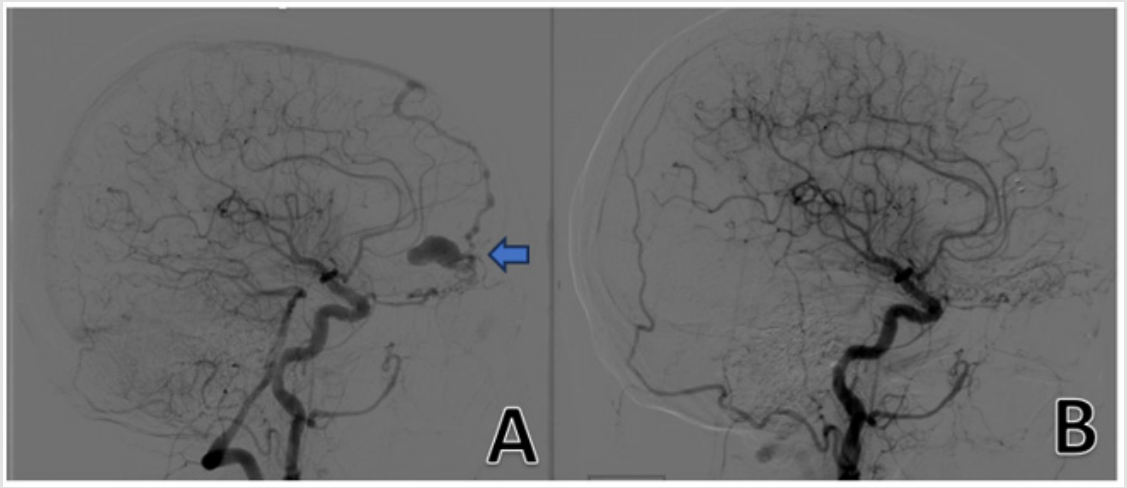

Figure 1: Case I:
A. Preoperative DSA showing frontobasal dural arteriovenous fistula, associated with giant venous aneurysm.
B. Postoperative angiography showing complete exclusion of the lesion.

Case II

A 50-year-old male presented to the emergency department, complaining about left facial numbness and paresthesia on the left side of the face. These symptoms were rapidly progressing. The patient was admitted to the neurosurgery department after a lesion was suspected in the cranial CT in the region of the pons. Significant associated edema was observed. The patient's condition deteriorated rapidly, with a decline in mental status and a GCS of less than 8. The patient was admitted to the ICU and intubated. A brain MRI was performed. A lesion in the pons was observed. The lesion itself appeared iso/hypodense on the T2 and FLAIR sequences (Figure 2A & 2C). A DSA was performed – a dural arteriovenous fistula with medial meningeal artery and meningohypophyseal trunk feeders, and associated with a giant venous aneurysm. (Figure 3A) The venous drainage appeared to be through peri-medullary veins and sinus rectus. There were several attempts to embolize the dural arteriovenous fistula with no success, justifying the decision to pursue surgical treatment. The patient underwent a retrosigmoid craniotomy and exclusion of the AVF. A DSA was performed after surgery and showed a complete exclusion of the dural AVM (Figure 3B). He was extubated on the second day and later discharged to a rehabilitation facility. A 6-month postoperative MRI showed no remnants of the lesion and complete resolution of the brainstem edema. (Figure 2B & 2D)

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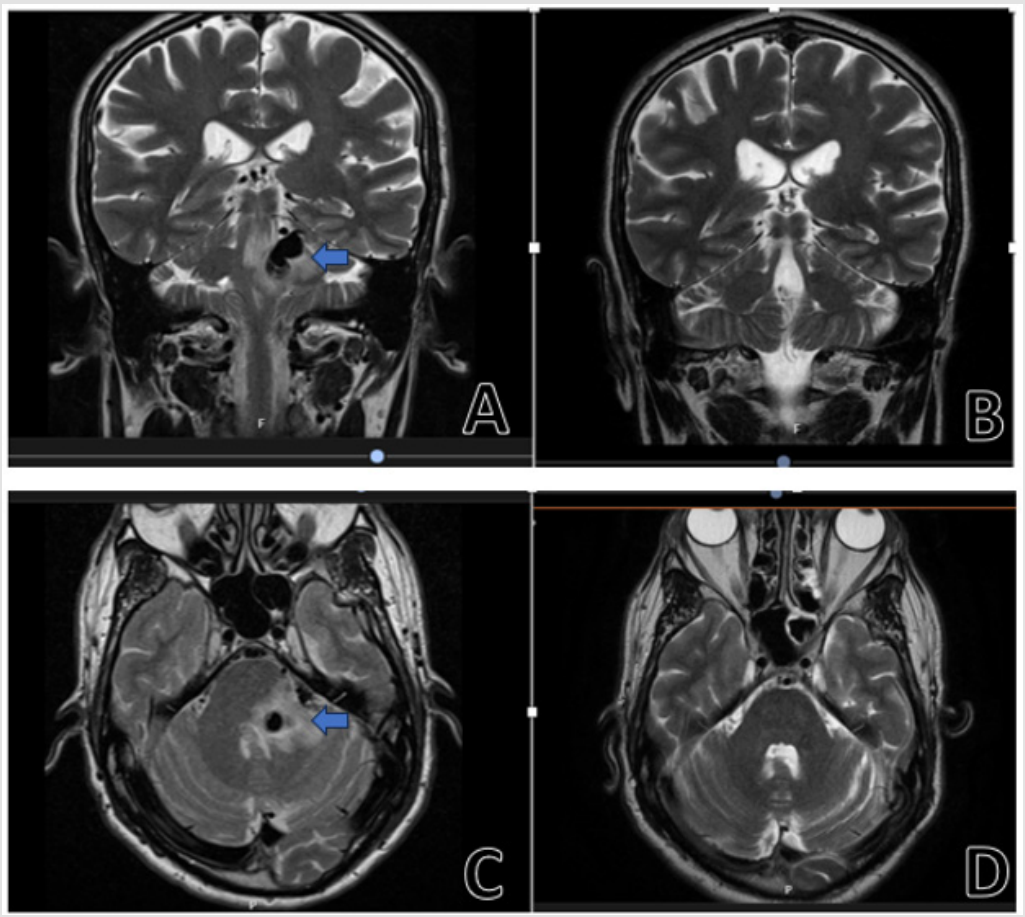

Figure 2: Case II

A- Coronal T2 MRI presents a dural arteriovenous fistula located in the pons, with associated edema.

B- B- Postoperative MRI showing exclusion of the lesion. C- Axial FLAIR preoperative MRI showing the previously described lesion. D- Axial T2 postoperative MRT showing the exclusion of the lesion).

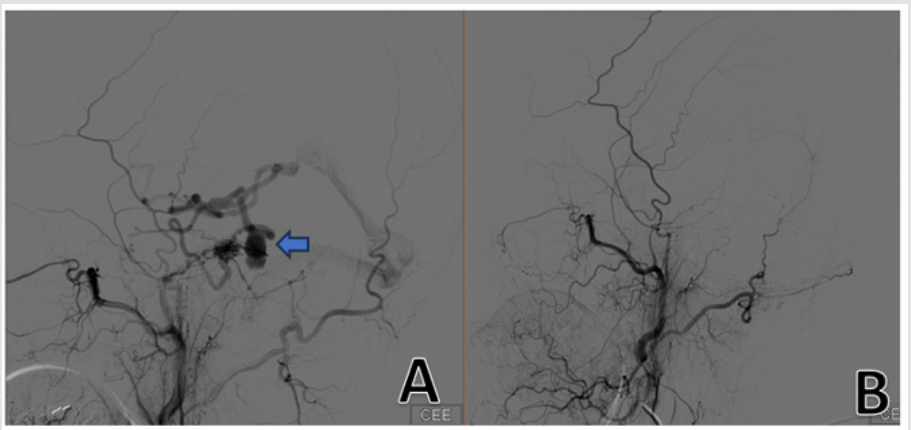

Figure 3: Case II:

A - Preoperative DSA showing dural arteriovenous fistula with medial meningeal artery and meningohypophyseal trunk feeders and associated with a giant venous aneurysm.

B - Postoperative DSA showing complete exclusion of the lesion).

Discussion

Intracranial dural arteriovenous fistulas, also known as dural arteriovenous shunts, are pathological connections between dural and pial arteries and veins within the dura mater, comprising the walls of the dural sinuses, bone traversing emissary veins, and bridging venous structures [1]. These lesions were first described by Razzoli in 1881, and the first angiographic evidence of this disease was published by Sachs in 1931. Dural arteriovenous fistulas are considered a rare pathology, accounting for 10-15% of all intracranial vascular malformations [2]. However, cases of selected lesions remaining fully asymptomatic and/or resolving spontaneously have been reported in the literature; therefore, their incidence might be underreported [3]. Dural arteriovenous fistulas are most often located near the transverse and cavernous sinuses, but can occur at any location within the intracranial dura mater [4]. The current treatment modalities go from conservative management to endovascular embolization, stereotactic radiosurgery, and microsurgical resection.

Endovascular Treatment

Management of dural arteriovenous fistulas using endovascular embolization is mainly considered first-line treatment for most of these lesions. The techniques used for embolization include trans–arterial, transvenous, and direct puncture, which is used infrequently [5]. In the case of trans-arterial embolization, the obliteration of feeding branches aims at reducing blood flow to the shunt but can be limited by difficulties in catheterizing feeding arteries and compromising the complete obliteration of the dural arteriovenous fistulas. As such, the trans arterial embolization method can usually be applied as an adjunct treatment to stabilize the dural arteriovenous fistulas before other procedures (surgery or radiotherapy) are attempted [5]. The most recently used embolization method has become transvenous embolization due to its association with higher rates of complete arteriovenous fistula obliteration. This method is most prominently utilized in treating dural arteriovenous fistulas of the cavernous, sigmoid, and transverse sinuses [1,5,6]. Various materials can be used for dural arteriovenous fistula embolization. In the case of the trans-arterial route, liquid embolic agents are commonly used, like Onyx, PHIL, or Squid. In the transvenous approach, coiling of the draining veins or their occlusion with liquid embolization agents is the preferred method [7-9].

Surgical Treatment

Despite being used less frequently due to the advancement of endovascular techniques, surgical treatment remains an effective alternative for the treatment of dural arteriovenous fistulas. It can be chosen when the patient presents with a hemorrhage that requires evacuation or serious neurological deficits, such as seizures or paresis [10], which was the case for more than half of our patient group. Open surgical treatment also holds a role in treating fistulas that are difficult to access endovascularly, or those that cannot be conveniently accessed endovascularly [11,12], and therefore it is also a mainstay treatment for ethmoidal dural arteriovenous fistulas. Some dural arteriovenous fistulas are located in the anterior cranial fossa. For these lesions, surgical treatment is associated with far superior results in both obliteration rates and patient outcomes [13]. Also, open surgical treatment can be used in reserved cases where endovascular approaches have previously failed to completely obliterate the dural arteriovenous fistula [14]. In the case of a dural arteriovenous fistula of the transverse-sigmoid sinus region, the surgical procedure involves skeletonization of the involved sinus and the coagulation of dural feeding arteries and arterialized cortical veins [15]. In the case of dural arteriovenous fistulas without direct drainage into a dural sinus, the lesion is managed with the cortical draining vein being disconnected from the fistula point by bipolar coagulation or micro clips [16].

Stereotactic Radiosurgery

SRS is often reserved as the last treatment option for dural arteriovenous fistulas. This method is thought to induce endothelial cell damage and subsequent thrombosis of the fistula, which leads to occlusion of the lesion [17]. Similarly with the treatment of cerebral AVMs, the obliteration of the dural arteriovenous fistula can take months, during which time the risk of bleeding is still present [18,19]. More benign Cognard grade I or Borden grade I dural arteriovenous fistulas are considered good candidates for treatment with SRS. These lesions are associated with a higher rate of obliteration without bleeding during the post-treatment latency period [18]. Stereotactic radiosurgery is also an option for high-grade arteriovenous fistulas when surgical or endovascular treatment approaches have failed to achieve obliteration or are deemed too dangerous to perform [17]. Treatment with SRS, according to the literature, can produce a complete obliteration rate from 50 to 93% [17, 18, 20]. However, the average latency period of dural arteriovenous fistula closure is reported as 23 months after the procedure, and the annual rebleeding rate is reported as high as 2.6% [20,21]. Therefore, even though effective, this treatment method carries a significant risk of hemorrhagic events in susceptible cases. We do not present any patients treated with this method in our cohort [22-24].

Conclusions

Even though surgical treatment is nowadays less frequently used, it still remains a viable option for the treatment of dural arteriovenous fistulas. Our patient cohort demonstrated that selectively treating these rare and complex lesions surgically is associated with high obliteration rates and relatively low rates of complications. Surgical treatment can be considered a primary treatment option when dealing with ethmoidal-type dural arteriovenous fistulas or others located within the anterior cranial fossa. Treating these lesions surgically is also a viable option when a patient presents with severe neurological symptoms and/or hemorrhage that requires evacuation, as well

as when endovascular embolization methods have failed. Multidisciplinary team discussion should remain the focus of adequate decision-making on an individual-patient basis.

Limitations of the Study

This study has several limitations. The biggest of which is its retrospective nature and small sample size. To accurately determine one method's superiority over another, the clinical study should be prospective in its nature. The lack of patient randomization is also a limiting factor. No stereotactic radiosurgery cases were included in this study since our center does not possess this treatment modality. Therefore, more prospective randomized trials should be conducted in the future in larger sample sizes to fully evaluate the optimal treatment method for various locations of dural arteriovenous fistulas.

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