

Management and Outcome of Intracranial Dural Arteriovenous Fistulas That Have Caused a Hemorrhage in the Posterior Fossa : A Clinical Study

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Objective : We evaluated the diagnosis, treatment, and long-term results of patients with dural arteriovenous fistula (dAVF), which is a very rare cause of posterior fossa hemorrhage.

Methods : This study included 15 patients who underwent endovascular, surgical, combined, or Gamma Knife treatments between 2012 and 2020. Demographics and clinical features, angiographic features, treatment modalities, and outcomes were analyzed.

Results : The mean age of the patients was 40±17 years (range, 17–68), and 68% were men (11/15). Seven of the patients (46.6%) were in the age group of 50 years and older. While the mean Glasgow coma scale was 11.5±3.9 (range, 4–15), 46.3% presented with headache and 53.7% had stupor/coma. Four patients (26.6%) had only cerebellar hematoma and headache. All dAVFs had cortical venous drainage. In 11 patients (73.3%), the fistula was located in the tentorium and was the most common localization. Three patients (20%) had transverse and sigmoid sinus localizations, while one patient (6.7%) had dAVF located in the foramen magnum. Eighteen sessions were performed on the patients during endovascular treatment. Sixteen sessions (88.8%) were performed with the transarterial (TA) route, one session (5.5%) with the transvenous (TV) route, and one session (5.5%) with the TA+TV route. Surgery was performed in two patients (14.2%). One patient (7.1%) passed away. While there were nine patients (64.2%) with a Rankin score between 0 and 2, the total closure rate was 69.2% in the first year of control angiograms.

Conclusion : In the differential diagnosis of posterior fossa hemorrhages, the differential diagnosis of dAVFs, which is a very rare entity, should be considered, even in the middle and elderly age groups, in patients presenting with good clinical status and pure hematoma. The treatment of such patients can be done safely and effectively in a multidisciplinary manner with a good understanding of pathological vascular anatomy and appropriate endovascular treatment approaches.

Key Words : Arteriovenous fistula · Hemorrhage · Endovascular procedure · Therapeutic embolization · Posterior fossa hemorrhages.

• Received : March 27, 2023 • Revised : May 25, 2023 • Accepted : June 7, 2023

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INTRODUCTION

Spontaneous posterior fossa hemorrhages are between 5% and 13% of all intracranial hemorrhages, and the most common cause is hypertensive vasculopathies. Due to its relatively narrow volume and hosting important structures, such as the brain stem and cranial nerves, morbidity rates can be as high as 49.7% and mortality rates can be as high as 66% in posterior fossa hemorrhages^{5,21,26,28}. Although posterior fossa hemorrhage due to vascular pathologies, such as intracranial dural arteriovenous fistula (dAVF), is quite rare, the annual risk of re-bleeding after the first hemorrhage is up to 46%, indicating the importance of differential diagnosis and appropriate treatment^{7,9}. dAVFs are pathological shunts between the meningeal and/or pial arteries and the dural sinus or meningeal veins. While they are seen in 10–15% of all intracranial arteriovenous malformations (AVMs), they are generally considered as acquired cerebrovascular pathologies^{8,12}.

The treatment of intracranial dAVFs becomes very difficult when the complex vascular nutrition originating from the external carotid artery (ECA), the internal carotid artery (ICA), and the vertebrobasilar system, as well as the frequent cerebellar cortical venous drainage, are added together with the bleeding presentation. This increases the importance of performing stepwise procedures with a multidisciplinary approach^{4,6,15,18,24}. With the development of endovascular techniques, the anatomical and pathophysiological features of the fistula have been better understood, and it has been possible to perform patient-based treatment with the highest closure rates and low risk. Closure of proximal venous drainage, which is the main goal of treatment, has been the main treatment management for most dAVFs, especially with the continuous development of embolic liquid agents, such as Onyx, which can achieve high proximal drainage vein penetration rates^{12,15,16}.

The current article explains the differential diagnosis, clinical presentation, treatment, and long-term angiographic and clinical outcomes of intracranial dAVFs that result in bleeding in the posterior fossa, with an emphasis on endovascular therapy (EVT).

MATERIALS AND METHODS

Between 2012 and 2020, 212 patients with AVF with intracranial localization who were treated in our neurosurgery clinics were retrospectively analyzed. Study guidelines and protocol was approved by Bursa Yüksek İhtisas Training and Research Hospital Ethics Committee (2011-KAEK-25 2021/08-16). The treatments were performed by an endovascular, surgical, or combined neurosurgeon and neuroradiologist as multidisciplinary. Patients who had bleeding in the posterior fossa were included in the study. The exclusion criteria were defined as patients with AVM bleeding, no long-term follow-up, and poor image quality. Fifteen patients were included in the study with these criteria. Age, gender, symptoms, Glasgow coma scale (GCS), the type of bleeding (subarachnoid hemorrhage [SAH], parenchymal, and intraventricular), fistula location, angiographic features, Cognard classifications³, feeders, venous drainage, treatment modalities (external ventricular drainage [EVD], EVT, surgery, Gamma Knife treatment [GKT]), clinical, and radiological follow-ups were examined. Fistulas were divided into dural and tentorial AVF. Fistulas located in the transverse and sigmoid sinus were considered as dAVF located in the dural. On the basis of sinus and venous drainage direction, dural-tentorial dAVFs were considered as tentorial dAVF (Galen, insular, petrosal, torcular, tentorial, and straight sinus localized)¹⁴. All cases were evaluated with digital subtraction angiography (DSA) (with bilateral ICA, ECA, and vertebral arteries) after examination with computerized tomography (CT), computerized tomography angiography (CTA), or magnetic resonance imaging (MRI), magnetic resonance angiography (MRA), and after the necessary initial medical, intensive care, and invasive (EVD) treatments. The primary treatment of patients evaluated as multidisciplinary by the neurovascular team at each center was EVT. This was achieved using transarterial (TA), transvenous (TV), or combined TA/TV endovascular approaches. Onyx (ev3, Irvine, CA, USA) and coil were preferred as embolization materials. In the presence of hemorrhage due to brain stem compression and hydrocephalus after EVT, surgery was performed to evacuate the hematoma via the suboccipital route and to remove the fistula. GKT was applied to a patient with dAVF (patient 11) who had partial closure after two sessions of EVT. Clinical discharge and 1 year of clinical status were categorized by modified Rankin scale (mRS) as good outcome (mRS

Table 1. Patients' demographic features, clinical presentation, angiographic findings, treatment modalities and outcomes

Patient No.	Age (years)	Sex	Clinical presentation and initial GCS score	Hemorrhage	Localisation	Feeders	Cognard classification	Treatment	Session	mRS	1st year mRS	Obliteration
1	50	M	Headache/15	SAH+parenchymal	Dural	Bilateral OA-PMA, MMA	IV	TA	1	2	0	Near total
2	37	M	Coma/6	SAH+parenchymal+intraventricular	Tentorial	MHT, OA-PMA	IV	EVD+TV	1	3	1	Total
3	20	F	Stupor/11	SAH+parenchymal	Tentorial	SCA, OA-PMA	III	EVD+TA	3	3	1	Total
4	51	M	Stupor/10	SAH+intraventricular	For. Magnum	VA-PMA, APA	IV	EVD+TA	1	4	3	Total
5	17	F	Headache/15	Parenchymal	Tentorial	SCA, OA-PMA	III	TA	1	3	1	Near total
6	22	M	Stupor/13	Parenchymal+intraventricular	Tentorial	SCA, MHT	IV	EVD+TA	1	3	2	Near total
7	35	M	Headache/15	SAH	Tentorial	MMA, OA-PMA, SCA	III	TA	1	3	1	Total
8	58	M	Headache/14	Parenchymal	Dural	MMA, OA-PMA, MHT, VA-PMA	Ila+b	TA+TV	2	2	1	Total
9	29	M	Headache/15	Intraventricular	Tentorial	MHT, SCA	III	Observation	0	2	1	
10	21	M	Coma/6	SAH+intraventricular	Tentorial	VA-PMA, OA-PMA, MMA	III	EVD+TA	1	6	6	Total
11	68	M	Headache/15	Parenchymal	Tentorial	MHT, MMA+OA-PMA	Ila+b	TA+GKT	2	1	1	Partial
12	29	F	Stupor/12	SAH+parenchymal	Tentorial	AICA, MMA	III	EVD+TA	1	3	2	Total
13	50	M	Headache/15	Parenchymal	Dural	Bilateral OA-PMA	IV	TA+surgery	1	3	1	Total
14	65	M	Coma/4	SAH+parenchymal+intraventricular	Tentorial	OA-PMA, MMA, VA-PMA	IV	EVD+TA+surgery	1	5	5	Total
15	61	M	Coma/7	SAH+intraventricular	Tentorial	SCA, MMA, MHT	III	EVD+TA	1	5	3	Total

GCS : Glasgow coma scale, mRS : modified Rankin scale, M : male, SAH : subarachnoid hemorrhage, OA : occipital artery, PMA : posterior meningeal artery, MMA : middle meningeal artery, TA : transarterial, MHT : meningohypophyseal trunk, EVD : external ventricular drainage, TV : transvenous, F : female, SCA : superior cerebellar artery, For : foramen, VA : vertebral artery, APA : anterior pharyngeal artery, GKT : Gamma Knife treatment, AICA : anterior inferior cerebellar artery

0–2) or poor outcome (mRS 3–6). First year of fistula closure rates were determined with the help of DSA, MRA, and CTA.

Statistical analysis was performed with SPSS ver. 26 (IBM, Armonk, NY, USA). Fisher's exact test, independent t-test and chi-square test of independence were the tests used for analysis. $p < 0.05$ was accepted as statistical significance. Percentages were used for categorical variables and mean \pm standard deviation for continuous variables.

RESULTS

The mean age of the patients was 40 ± 17 years (range, 17–68), and 68% were men (11/15). Seven of the patients (46.6%) were in the age group of 50 years and older. While the mean GCS was 11.5 ± 3.9 (range, 4–15), 46.3% presented with headache and 53.7% had stupor/coma. Cerebellar hematoma was the most common type of bleeding in 10 (66.7%) of posterior fossa hemorrhages detected in the first CT, SAH in nine (60%), and intraventricular hemorrhage in seven (46.7%) detected (Table 1). Four patients (26.6%) had only cerebellar hematoma and headache. Seven patients (46.7%) underwent

EVD due to acute hydrocephalus. DSA was performed in all patients within the first 24 hours after admission. According to the Cognard classification, six of the patients (40%) were type IV, seven (46.7%) were type III, and two (13.3%) were type IIa+b, and venous aneurysm was also observed in five patients (33.3%). Parenchymal hematoma was present in the vicinity of 80% of patients with venous aneurysm. All dAVFs had cortical venous drainage. In 11 patients (73.3%), the fistula was located in the tentorium and was the most common localization. Transverse-sigmoid sinus (TSS) localizations were three (20%), while dAVF located in the foramen magnum was detected in one patient (6.7%). The main dAVF feeding artery was the occipital artery (OA) and was detected in 12 patients (30.7%). Middle meningeal artery (MMA) eight (20.5%), superior cerebellar artery (SCA) six (15.3%), meningo hypophyseal trunk (MHT) six (15.3%), vertebral artery (VA) five (12.8%), anterior inferior cerebellar artery (AICA) and anterior pharyngeal artery one (2.5%) as dAVF feeders watched (Table 1). While the venous drainage of five patients (33%) was direct TSS, three patients (20%) were found to be draining to superior petrosal sinus (SPS), two patients (13.3%) to torcula, and two patients (13.3%) to basal vein. Venous drainage was

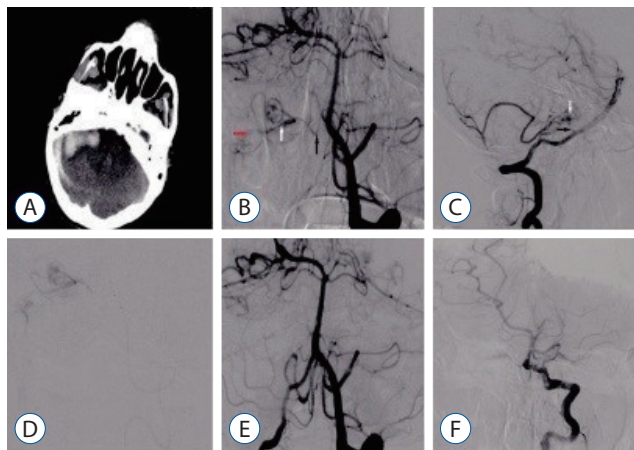


Fig. 1. A 29-year-old male patient (patient 12) was admitted to the hospital with confusion and vomiting. A : Right cerebellar hematoma on noncontrast cranial computed tomography. B and C : Left vertebral artery digital subtraction angiogram, in anterior-posterior and lateral projection, fed from the right pial supply from the AICA (black arrow), draining into the superior petrosal sinus (red arrow) via retrograde venous drainage superior petrosal tentorial dural arteriovenous fistula (white arrow). D : Embolization with Onyx after microcatheterization via AICA. E and F : Complete closure of the fistula in the post-operative first year of follow-up. In the first year of follow-up, it was observed that the sixth nerve paralysis continued. AICA : anterior inferior cerebellar artery.

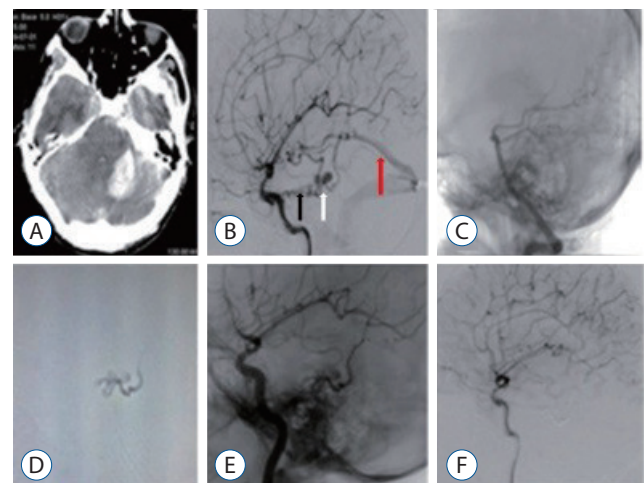


Fig. 2. A 37-year-old male patient (patient 2) in a coma after sudden headache. A : Left cerebellar hematoma, intraventricular hemorrhage, and subarachnoid hemorrhage were observed on noncontrast cranial computed tomography. B and C : Lateral left carotid and vertebral artery digital subtraction angiogram showed a tentorial dural arteriovenous fistula (white arrow) draining into the straight sinus with deep venous structures (red arrow), fed from the left meningo hypophyseal artery (black arrow). D and E : Onyx embolization by transvenous route. F : No residue was observed in the first year of follow-up.

observed in the perimedullary venous plexus, vein of Galen, and one (6.6%) each in the tentorial sinus. Eighteen sessions of EVT were applied to 14 patients, except for the dAVF patient with cerebellar peduncle localized intraventricular hematoma who did not accept treatment. dAVFs were closed with 16 sessions (88.8%) of TA route (Fig. 1), and one session (5.5%) with TV (Fig. 2) and TA+TV. A total of 20 feeder arteries were accessed via the TA route, and the most commonly preferred artery was MMA (45%) (Table 1). Emergency surgery was performed after EVT in two patients (14.2%) (patients 13 and 14) who had brain stem compression due to hematoma. As a procedure complication, a permanent sixth cranial nerve lesion was observed in one patient (patient 12). Of the 14 treated patients, one patient required a permanent ventriculoperitoneal shunt. One patient (7.1%) passed away. While the number of patients discharged between mRS 0 and 2 was three (21.4%), this number was observed to be nine (64.2%) in the first year. The first-year angiographic follow-ups of 14 patients were performed with DSA in 11 patients (78.5%) and with MRA and

CTA in the remaining three patients. In 10 patients (71%), three (21.4%) of the dAVF were found to be near total, and one (7.1%) was partially obliterated. One patient with partial closure was referred to GKT. Recurrence was observed in one patient in the sixth month control DSA (Fig. 3). During the second session TA-EVT was planned, the patient who became pregnant during the follow-up (total and near total) presented with cerebellar hematoma for the second time in the second trimester (12th month of the first bleeding), was followed up with EVD, did not accept further treatment. In the postpartum 12th month (32nd month of the first hemorrhage), the 2nd and 3rd sessions were closed completely by the TA route.

Statistically, the most defining feature for the discharge and 1-year favorable mRS (0–2) values were the high admission GCS score and the absence of intraventricular bleeding ($p < 0.05$).

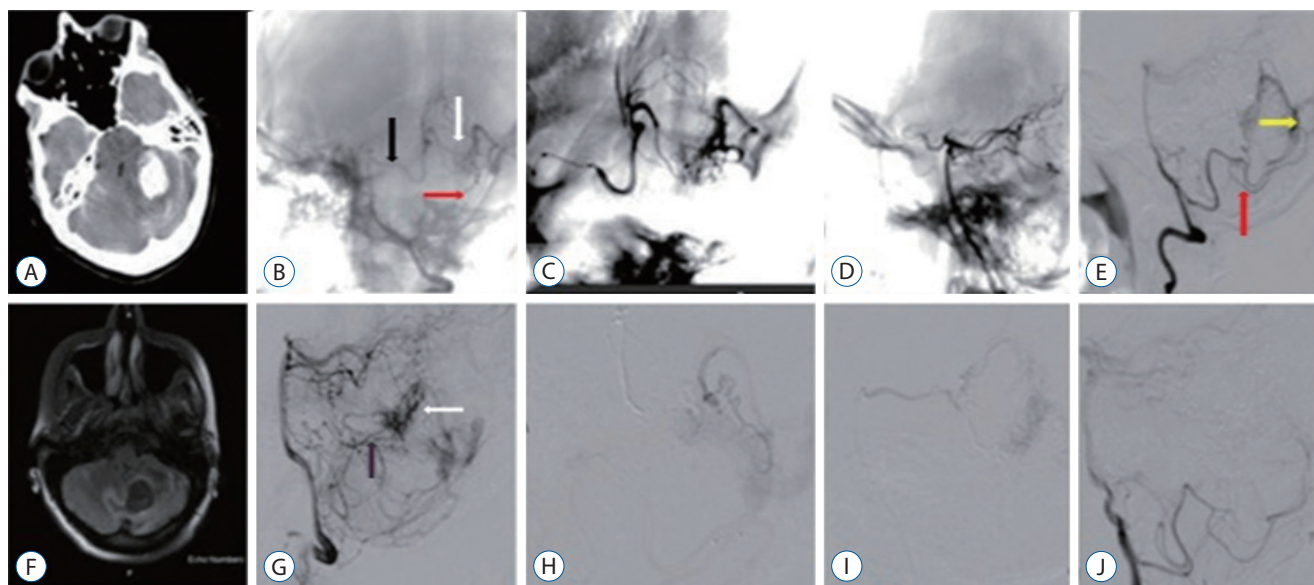


Fig. 3. A 20-year-old female patient (patient 3) is admitted to the hospital with sudden onset of vomiting, confusion, and left hemiparesis. A : Noncontrast cranial computed tomography showed left cerebellar hematoma, subarachnoid hemorrhage, and hydrocephalus (triventricular hydrocephalus not included in cross-sections). B : In the left vertebral artery digital subtraction angiogram, a tentorial dural arteriovenous fistula (white arrow), which is fed via the left superior cerebellar artery (black arrow) and VA-PMA (red arrow) draining into the torcula (yellow arrow) via the retrograde cortical venous pathway, is observed. C and D : Onyx embolization by catheterizing the left superior cerebellar artery using the transarterial route. E : At the sixth month of follow-up, it was observed that the fistula was reopened via VA-PMA. Second session is recommended. The patient refused because she was pregnant. F : Cerebellar hematoma and hydrocephalus were observed in magnetic resonance imaging performed after unconsciousness in the second trimester. The general condition improved with external ventricular drainage, but the patient refused farther treatment. There was no need for a permanent ventriculoperitoneal shunt. G : In the DSA performed in the first year of postpartum, the tentorial fistula draining into the torcula, in which the main originating arteries are superior cerebellar artery and VA-PMA, was completely closed in two different sessions (H and I), in three sessions in total. J : First-year control DSA. VA-PMA : vertebral artery-posterior menegial artery, DSA : digital subtraction angiography.

DISCUSSION

Hypertensive cerebellar hemorrhages are mostly seen in the middle and elderly group and are often thought to originate from dentate nucleus localization. Bleeding from this region usually tends to open into the fourth ventricle in addition to the presence of a large hematoma, but the presence of SAH is not a typical finding^{5,17}. Because they rarely cause infratentorial hemorrhage, the diagnosis can be delayed, which can lead to high rates of secondary hemorrhages^{9,24}. Posterior fossa hemorrhages due to dAVFs tend to be small because they are of venous origin, although they are arterialized^{4,13}. It is often accompanied by intraventricular and/or SAH and a severe decrease in consciousness²². While there were CT and clinical findings suggestive of dAVF bleeding at the first admission of 11 (73.3%) posterior fossa bleeding in our series, it was observed that four patients (26%) presenting with pure cerebellar hematoma presented clinically only with headache. In addition, seven of the patients (46.6%) in our series consisted of the age group of 50 years and older, who could be diagnosed with cerebellar hematoma due to hypertensive vasculopathy as a preliminary diagnosis. It was observed that approximately half of these patients presented only with headache and pure cerebellar hematoma. While suggesting that these clinical and radiological findings in our study may be important parameters showing the importance of the differential diagnosis, especially with hypertensive cerebellar hemorrhages, it also reminds us of the importance of a multidisciplinary approach.

The main purpose of all intracranial AVF treatment is to separate the venous structure, which is the drainage foot of the fistula, and to completely close the proximal venous drainage, and only in this way can the treatment be terminated^{4,15,19,24}.

There are many studies in the literature on the surgical approach based on principles, such as separation of the fistula with venous drainage, excision of the lesion, and closure of the drainage sinus, especially since the early 1980s, and low recurrence rates have been reported with high total closure rates^{4,20,25}. However, surgery of infratentorial dAVF lesions, which have a narrow surgical field, edematous cerebellum parenchyma, difficulty in recognizing normal/abnormal drainage veins, full drainage veins, and complex vascular structures presenting with bleeding, in addition to adjacent to important structures, is quite difficult and may have high morbidity

rates. Surgery was not the first choice in our study, and it was applied to two patients who needed decompression due to brain stem compression of the hematoma after EVT.

Because of the rarity of dAVFs that have caused bleeding in the posterior fossa, their endovascular treatment is less well-defined^{6,18}. In our series and EVT approach preference was determined according to fistula localization and venous outflow. Although the most effective options seem to be the TV or combined EVT approach, the most common approach is TA¹⁵. As in many previous studies, our first choice (88.8%) was the TA approach. The most preferred artery was MMA (45%), even though it is not the main feeder, because it contains less tortuosity and has low-hazard anastomotic structures. Embolization through the meningeal branch of the SCA was the first option in fistulas with tentorial localization at a level where distal access is possible and Onyx reflux is acceptable (25%). In a Foramen magnum dAVF patient with perimedullary venous drainage, where TV access was not possible, the direct TA route was the reason for preference, and the fistula was closed via a PMA feeder (patient 4). In a patient with tentorium localization where MHT is the main feeder, direct straight sinus access and TV route was preferred due to the risk of difficult access and reflux in the ICA (Fig. 2). In a patient who could not achieve adequate closure by TA, we preferred a combined (MMA+TSS) treatment approach in the second session (patient 8). In a patient with tentorium dAVF with pial supply from the AICA and venous outflow to the SPS, the TA approach was preferred because distal access via AICA was possible (Fig. 1). With this EVT strategy, we have implemented in our series, it has been possible to achieve high closure rates with low risk.

Peto et al.¹⁸ noted the clinical results of 28 patients with posterior fossa dAVF, nine (32.1%) of whom presented with bleeding who were treated with surgery, EVT, GKT, or a combination of multiple modalities. In the series, most of the dAVFs (82%) were reported to be completely closed; their recurrence rate was 13%; one patient passed away, and 25% required surgery. GKT was applied in one patient because closure could not be achieved despite repeated EVT treatments¹⁸. In a recent dAVF series of 26 cases with posterior fossa localization, it was reported that eight patients (30%) presented with bleeding, and 23 patients (89%) underwent EVT via TA⁶. While the complication rates related to the procedure were 15%, it was reported that the complications were completely

resolved in the sixth month follow-up, and the sixth month closure rate was 100%. While the first-year total closure rate of the patients treated in our series was 71%, near total closure was 21.7%. Recurrence was observed in one patient, while mRS at first year (0–2) was 64.2%. Bleeding due to dAVFs appears to have a more benign clinical course compared with bleeding from an aneurysm⁶⁾. Our clinical results lagged behind two other similar studies, the fact that our series consisted of only bleeding patients and can be attributed to the natural course of posterior fossa hemorrhages, which already have high morbidity and mortality rates. The bad effects of presentation with low GCS and the presence of intraventricular bleeding on clinical outcomes were also shown statistically ($p < 0.05$). However, our long-term clinical and radiological results were found to be similar when compared with the case series of bleeding intracranial AVF^{4,15)}.

The risk of bleeding continues because GKT, which aims to obliterate the fistula by vascular thrombosis, needs a latent period that can extend to 2 years. Therefore, it is not suitable because it is known that the risk of rebleeding is high in bleeding dAVFs¹⁰⁾. This technique should be reserved for acceptable dAVFs, for which options are exhausted. In our study, GKT treatment was applied to one patient (patient 11) who was embolized twice, and partial closure was achieved. Although there was no obvious closure in the first year of follow-up, there was no hemorrhagic clinical.

Studies have shown that dAVF hemorrhages are mostly inside the parenchyma. It can also be accepted that there is a venous drainage system that has invaded parenchymal structures, is long, enlarged, and often contains ectasia and aneurysmal structures^{4,11)}. In our cases, the most common bleeding pattern was parenchymal hematoma, and 80% of them were accompanied by venous aneurysm.

In another large series, the most common bleeding localizations were shown as TSS, tentorium, and convexity, and in our study, the bleeding localizations appeared to be adjacent to the tentorium and TTS²³⁾. Tentorial dAVF fistula (66.7%) was the most common in this series. AVFs in this localization frequently have direct drainage into cortical veins; patent sinus drainages are rare and often associated with venous ectasia formation (Cognard IV). These features make them prone to intracranial bleeding. Although there are microsurgery series reporting high closure rates and low complication rates^{2,14,24)}. EVT is the most frequently applied approach, which

is less invasive and reported similar closure and complication rates with higher number of cases^{1,12,27)}.

The complication related to the procedure was the sixth CS palsy in one patient (7.1%) in whom AICA was used as the TA route (Fig. 1). In addition, it was seen that he persisted in the 1-year controls. In order to avoid preoperative complications in EVT, it reminds the importance of awareness of anatomical borders and the possibility of occlusion of cranial nerve feeders with embolizing agents, considering the presence of potentially dangerous anastomoses. Secondary hemorrhage due to recurrence (Fig. 3) was observed in one patient (patient 3), and a permanent ventriculoperitoneal shunt was required in one patient.

CONCLUSION

Since dAVFs that cause bleeding in the posterior fossa are very rare, secondary hemorrhages may be seen due to the possibility of a delayed or misdiagnosed diagnosis. In the differential diagnosis of cerebellar hematoma, even in the middle and elderly age groups, patients presenting with mild clinical findings and pure cerebellar hematoma should be evaluated multidisciplinary in terms of dAVF. In addition, good clinical results and high closure rates can be obtained because of a good understanding of pathological vascular anatomy and the application of appropriate patient-based treatments. Although it is retrospective and carried out with a small patient population, the results of our study may contribute to the handling of these lesions and assist in the larger multicenter series needed in this direction.

AUTHORS' DECLARATION

Conflicts of interest

No potential conflict of interest relevant to this article was reported.

Informed consent

This type of study does not require informed consent.

Author contributions

Conceptualization : RA; Data curation : RA, US, ED, İA,

VA; Formal analysis : RA, ADB; Methodology : RA, ED; Project administration : ED; Visualization : US, ED; Writing - original draft : RA, ED, İA; Writing - review & editing : ADB, ED, RA

Data sharing

None

Preprint

None

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