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REVIEW

Endovascular and surgical approaches of ethmoidal dural fistulas: a multicenter experience and a literature review

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Abstract Ethmoidal dural arteriovenous fistulae are rare vascular malformations associated with a high risk of bleeding. We present a multicenter contemporary series of patients treated with microsurgical and endovascular techniques. Sixteen consecutive patients were evaluated and/or treated between 2008 and 2015 at four centers with large experience in the endovascular and surgical treatment of cerebrovascular diseases. We analyzed demographic and clinical data, risk factors for dural fistulas, treatment type, peri- and post-operative morbidity, clinical and radiological outcomes, rates of occlusion, and long-term neurological outcome. Sixteen patients (81 % men, mean age of 58 years) with ethmoidal dural fistulas were included in the analysis. Seven patients had suffered an intracranial hemorrhage; the remaining presenting with neurological signs and symptoms or the fistula was an incidental finding. Three patients were managed conservatively. Among patients who underwent intervention (n = 13), 46.1 % were treated with endovascular therapy and 53.9 % were treated surgically. Complete angiographic obliteration was achieved in 100 % immediately after treatment and at last follow-up evaluation. All patients experienced a favorable neurological recovery (mRS 0–2) at the last follow-up visit (12 months). Ethmoidal dural AVFs are found mostly in male patients. Nowadays, due to wider use of non-invasive imaging, AVFs are discovered with increasing frequency in patients with minimal or no symptoms. Traditionally, these fistulas were

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considered "surgical." However, in the modern endovascular era, selected patients can be effectively and safely treated with embolization although surgical ligation continues to have an important role in their management.

Keywords Ethmoidal arteriovenous fistula · Endovascular treatment · Surgical disconnection · Arteriovenous malformation

Introduction

Intracranial dural arteriovenous fistulae (dAVF) account for 15 % of all cerebral vascular malformations [1], and only 10 % of all dAVFs are located in the anterior cranial fossa [2]. The ethmoidal artery is the main arterial vessel involved in the development of dAVFs of the anterior cranial fossa, which are more commonly indicated as ethmoidal dAVFs (e-dAVFs) [3]. The venous drainage of e-dAVFs involves, in most cases, frontal cortical veins, with retrograde drainage into the superior sagittal sinus with a tendency to the development of venous varicosities [4]. Because of their angioarchitecture, e-dAVFs are associated with a significant bleeding risk [5] and with a high risk of rebleeding soon after a presenting hemorrhage [6].

Similarly to dAVFs in other locations, the characteristics of the venous drainage of e-dAVFs are the main factor affecting clinical presentation (headache, seizures, and visual disorders), natural history, and choice of treatment [7]. Over the years, various therapeutic modalities have been proposed for the treatment of these fistulae: surgical disconnection, endovascular embolization, and radiosurgery as the sole therapy or in various combinations [8, 9] Because of the relative rarity of e-dAVFs, there are no large contemporary series available. In the present study, we describe a multicenter series of e-dAVF treated over a relatively short period of time with contemporary microsurgical and endovascular techniques(Figs. 1, 2, 3, 4, and 5).

Materials and methods

We retrospectively analyzed a consecutive series of patients with e-dAVFs treated between 2008 and 2015 at four tertiary referral centers specialized in the treatment of neurovascular disorders. Data analyzed included the following: age, gender, clinical presentation (hemorrhage, neurological symptoms, and incidental finding), risk factors predisposing to the development of arteriovenous fistulae (inherited or acquired coagulation abnormalities, intracranial tumors, history of head injury, pulmonary embolism and/or deep vein thrombosis), type of treatment, periprocedural complications, as well as clinical and radiological outcomes immediately after intervention and during follow-up (12 month). A complete catheter angiography was done to confirm occlusion of the fistula after treatment in most cases, while three patients were evaluated with magnetic resonance imaging angiography. Only one of the centers included in this study considered careful as a post-treatment radiological assessment, MRA. After diagnosis, all patients were evaluated by a team consisting of neurosurgeons and neurointerventional specialists to identify the best therapeutic strategy. The endovascular approach was considered as the first therapeutic option in each case; surgical treatment has been reserved for patients with major brain hemorrhage for cases in which the endovascular approach had failed. Three patients with asymptomatic e-dAVF have refused treatment and were managed conservatively. Patients with incidental

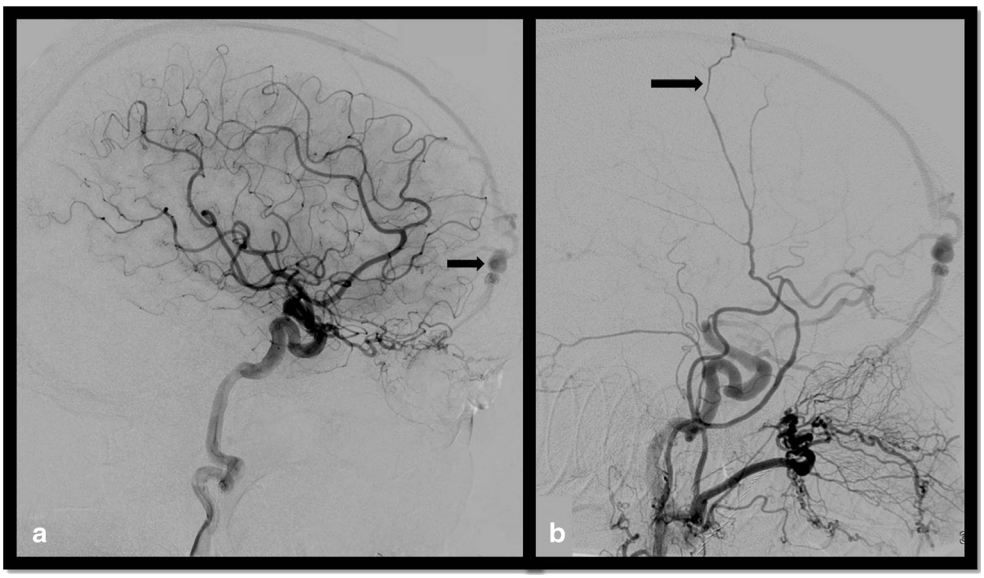

Fig. 1 An 80-year-old man presented with head "pressure" and was found to have an ethmoidal dAVF. **a** Catheter angiography, selective left internal carotid artery (ICA) injection shows ethmoidal branches of the ophthalmic artery feeding the fistula which drains through anterior ethmoidal veins to the superior sagittal sinus. There is a vein varix (*arrow*) associated with the venous drainage. **b** Left common carotid artery injection shows an enlarged branch of the middle meningeal artery also supplying the fistula

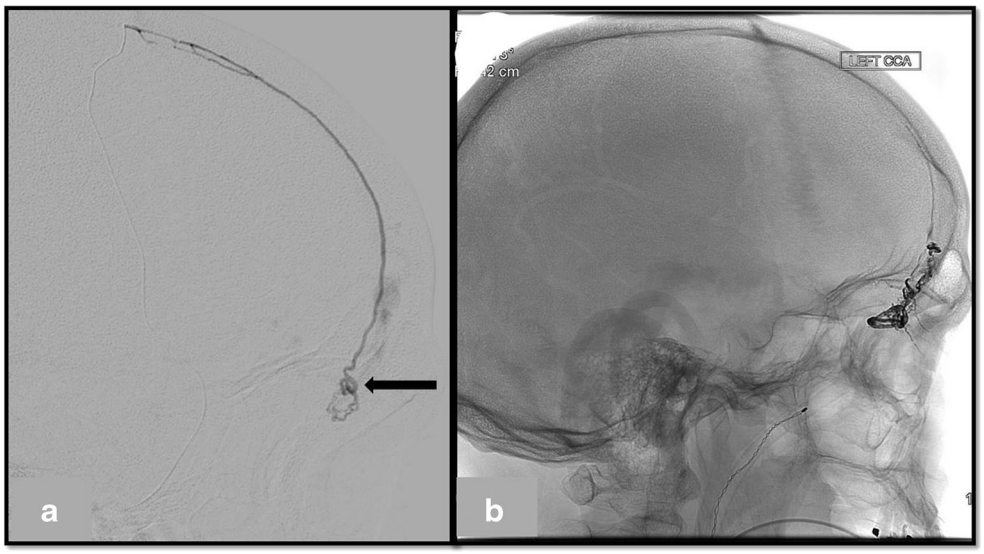

Fig. 2 Same patient in Fig. 1. **a** After selective catheterization of the distal middle meningeal artery, microcatheter injection shows supply to the intradural nidus (*arrow*) of the ethmoidal AVF. **b** Onyx cast after injection through the microcatheter in the distal middle meningeal artery in wedged position

lesions were treated conservatively because of advanced age and/or associated comorbidities (for example, one patient in whom the diagnosis of an e-dAVF was made during an angiogram for evaluation and treatment of acute stroke) and in absence of ominous angioarchitecture findings (i.e., venous varices and signs of outflow obstruction). Treatment of asymptomatic, incidental lesions was considered in those cases without the above mentioned factors.

Clinical assessment was evaluated by extracting the modified Rankin scale (mRS) score at each follow-up visit; poor outcome was defined as a mRS score ≥3.

Results

Sixteen patients with e-dAVFs were included in this series. Thirteen patients were male and the mean age was 58 years (range, 41–71 years). Six patients had suffered hemorrhage (3 presenting with sudden headache only, the others suffering from seizures, left-sided paresthesia, and coma). Three patients presented with symptoms other than hemorrhage: bruit (1 patient), proptosis (1 patient), and cognitive impairment (1 patient) while in seven patients the fistula was an incidental finding during radiological investigations done for various complaints not related to the e-dAVF. In our series, two patients presented risk factors associated with dAVFs: the first a pulmonary embolism in previous cerebral meningioma and the second one an aneurysm of the anterior communicating artery. Six-vessel digital subtraction angiography was performed in all cases. Clinical characteristics are summarized in Table 1.

Seven patients were treated with surgical disconnection, six underwent endovascular treatment, and three patients were managed conservatively. The combined approach is an option, but it was not performed in our series, since we consider e-dAVFs are usually relatively simple lesions to treat and therefore combined approaches, although described in the literature

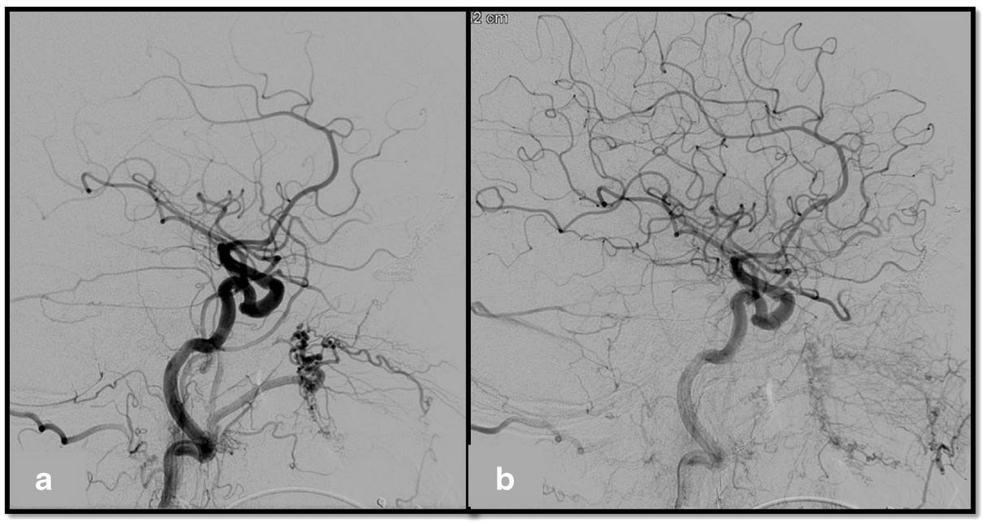

Fig. 3 Post-embolization left common carotid artery injection, early (**a**) and late (**b**) arterial phase shows complete obliteration of the fistula. The procedure was uneventful and the patient was discharged the following day

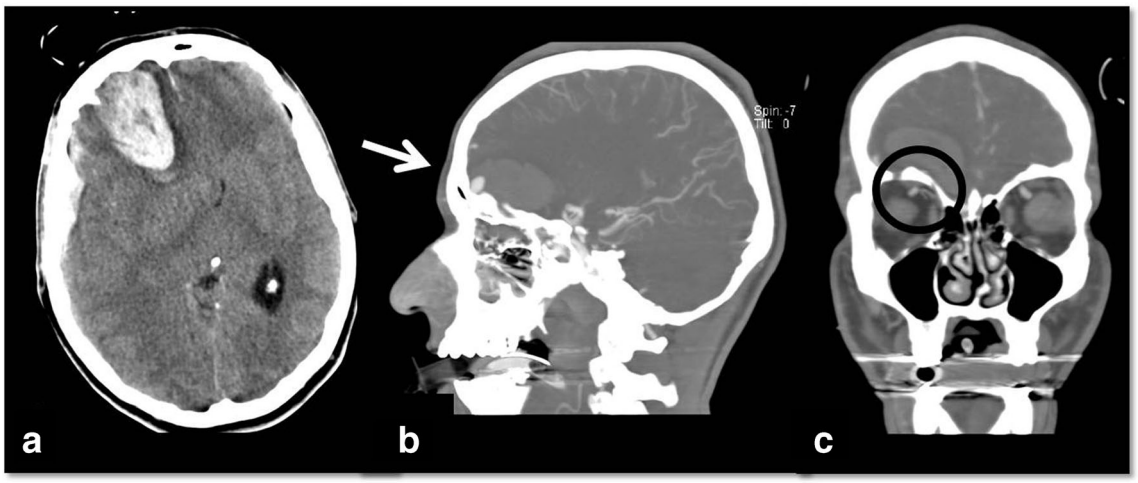

Fig. 4 A 52-year-old woman was admitted to a hospital with GCS 4 without traumatic onset. A CT scan showed a **a** right frontal hematoma with considerable midline shift. She underwent urgent CT angiography: sagittal view (**b**) showed a large vein varix (*white arrow*) associated with the venous drainage and coronal view (**c**) the wide ophthalmic artery (*black circle*)

(see Table 2) are rarely if ever necessary. Surgery was performed through a basal frontal approach, and the proximal portion of the draining vein was interrupted by placing a clip or by coagulating and dividing the proximal draining vein as it emerges from the dura. Post-operative angiographic obliteration of the e-dAVF was demonstrated in all of the seven patients (100 %) who underwent surgical treatment.

Endovascular procedures were performed under general anesthesia and systemic anticoagulation. The treatment consisted of embolization of the fistula and obliteration of the proximal portion of the draining vein with cyanoacrylate glue (Glubran, GEM, Viareggio, Italy), coils or Onyx (Covidien Neurovascular, Irvine, CA, USA). In four patients, a transarterial approach was utilized while in the remaining two patients the fistula was obliterated using a transvenous route.

Complications included a post-operative intraventricular hemorrhage in one patient treated with surgery (24 h after surgery) and a frontal hemorrhage in a patient undergoing endovascular treatment (48 h after treatment); both hemorrhagic complications were managed conservatively. The frontal hemorrhage resulted in slight right-sided weakness which persisted at follow-up while the patient with intraventricular hemorrhage did not suffer any permanent deficit. One patient treated with surgery suffered visual impairment from occipital ischemia secondary to an acute subdural hematoma occurring at presentation from rupture of the fistula. This patient

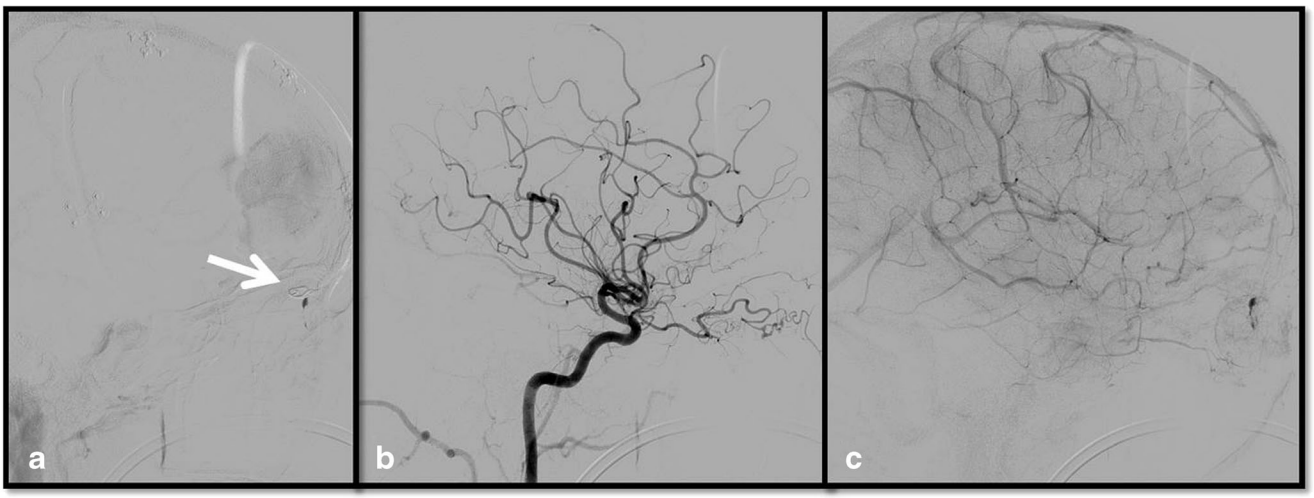

Fig. 5 A fast frontotemporal approach was performed in order to decrease intracranial pressure. A surgical disconnection of the ethmoidal fistula was obtained with clipping (**a**). A catheter angiography post-operative showed the correct placement of clip, and right common carotid artery injection, early (**b**) and late (**c**) arterial phase confirmed fistula resolution. The patient presented a visual impairment from occipital ischemia secondary to hemorrhagic onset. This patient experienced partial resolution of his visual deficit at 6 months

Table 1 Clinical characteristics

Number	M/F	Age	CVD	Hemorrhage	Sinus thrombosis	1st symptom	Treatment	Mode	Risk factors	Complications	Outcome radiographic	mRS
1	M	47	Y	N	N	Incidental	Surgery	Clip	PE/meningioma	None	C. O	0
2	F	69	N	N	N	Incidental	Conservative	–	N	None	–	–
3	M	68	Y	N	N	Incidental	Conservative	–	N	None	–	–
4	M	41	Y	Y	N	Headache	Endovascular	Onyx (transarterial approach)	N	None	C. O	2
5	F	52	Y	Y	N	Coma (GCS4)	Surgery	Clip	N	Visual deficit (post occipital ischemia)	C. O	1
6	M	48	Y	N	N	Incidental	Conservative	.	N	None	–	–
7	M	52	Y	N	N	Incidental	Surgery	Clip	N	None	C. O	0
8	M	65	Y	Y	N	Headache	Surgery	Clip	N	None	C. O	0
9	M	57	Y	Y	N	Headache	Surgery	Clip	N	None	C. O	0
10	M	72	Y	N	N	Cognitive impairment	Surgery	Clip	N	Hemorrhage	C. O	0
11	M	43	Y	N	N	Incidental	Endovascular	Coils (transvenous approach)	N	None	C. O	0
12	M	54	Y	N	N	Left bruit	Endovascular	Onyx (transarterial approach)	N	None	C. O	0
13	M	65	Y	Y	N	Left paresthesia	Endovascular	Coils (transvenous approach)	N	None	C. O	0
14	M	52	Y	Y	N	Seizures	Surgery	Clip	N	None	C. O	0
15	M	69	Y	N	N	Incidental	Endovascular	Onyx (transarterial approach)	N	Hemorrhage	C. O	1
16	F	71	N	N	N	Right proptosis	Endovascular	Onyx (transarterial approach)	ACoA aneurysm	None	C. O	0

experienced partial resolution of his visual deficit at 6 months. At the end of the 6-month follow-up interval, ten patients had a mRS of 0 (6 in the surgical and 4 in the endovascular group, respectively). Two patients had a mRS of 1: one patient treated with surgery who had presented with deep coma after hemorrhage from the fistula and another patient treated with endovascular embolization who was left with slight right-sided weakness. One patient treated with endovascular techniques had a mRS of 2 secondary to the effects of the initial presenting large hemorrhage. None of the patients managed conservatively experienced a hemorrhage during a follow-up period of 12 months.

Discussion

In a modern series of 16 patients with e-dAVFs managed at four centers over the past 8 years, we found a high male predominance (13/16 or 81 %) and most patients (10/16 or 62.5 %) presented with symptoms other than hemorrhage or harbored incidental lesions. Of treated patients, 46 % underwent successful endovascular treatment and overall outcomes were excellent with all but one patient having a modified Rankin score of 0–1 at follow-up.

The drastic male predominance of patients with e-dAVF, similar to the one observed in the case of tentorial dAVFs, is in net contrast with the clear female predominance observed in dAVFs involving the transverse/sigmoid sinus and the cavernous sinus [7]. The observation of definite different sex predilection in relation to the location of the dAVF led Lasjaunias and coworkers [22] to suggest that the three main dural compartments (ventral, lateral, and dorsal) share different biological, anatomical, and embryological characteristics which are reflected in the clinical characteristics of the dAVFs affecting these compartments. The anatomical features (associated bone, vascular, and parenchymal structures) of the three dural compartments, as well as the different characteristics of embryological development of each, render the vascular structures running through the dural sheaths of each compartment susceptible to certain risk factors able to facilitate the development of vascular anomalies such as dAVFs. These considerations would explain the clear predominance of females among patients with dAVFs of the ventral dural compartment and a clear predominance of males among those with dAVFs of the lateral compartment (which includes the anterior cranial fossa), due to different distributions of the frequencies of risk factors for the development of dAVFs in the two sexes [1].

In our series, the majority of patients (62.5 %) presented with symptoms other than hemorrhage. In 1990, Awad and collaborators [23] compiled of an extensive review of the literature on intracranial dural fistulas and reported that over 68 % of patients with anterior cranial fossa dAVFs reported up to that time had presented with “aggressive” neurological symptoms (the vast majority representing hemorrhage) [8]. Although we cannot exclude a referral bias, our observation that, in a modern series, most patients with e-dAVFs present with symptoms

Table 2 Cases of ethmoidal dural arteriovenous fistulas (literature review)

Author	Patients	Endovascular	Surgical	Conservative	Combined	Results
Deng [10]	5	5				Total obliteration
Spiotta [11]	3	3				Total obliteration
Mack [12]	2	2				Total obliteration
Abrahams [13]	7	1				Partial obliteration
			3			Total obliteration
				1		Total obliteration
					2	Total obliteration
Lawaton [14]	16		13			1 partial obliteration
						12 total obliteration
					3	Total obliteration
Hashimoto [15]	1		1			Total obliteration
Hattori [16]	1		1			Total obliteration
Ishikawa [17]	1		1			Total obliteration
Kikuchi [18]	2		2			Total obliteration
Baskaya [19]	2					
			2			Total obliteration
Martin [20]	8		6			Total obliteration
Halbach [21]	8		7			Total obliteration
				1		

other than hemorrhage may indicate a "shift" in the clinical presentation and diagnosis of e-dAVFs due to increased utilization of non-invasive diagnostic imaging studies and increased awareness. These factors may have resulted over time in a higher number of lesions being detected without hemorrhage. This observation is similar to the findings of changing clinical presentation over time in patients with tentorial dural AVFs [9, 24].

We summarized main cases of fistula reported in the literature in the Table 2; 56 cases were treated between 1990 and 2014, and in 11 cases an endovascular approach was applied. Thirty-six patients were treated surgically, two conservatively. And in five cases a combined approach was used. In two cases (1 treated with endovascular technique and 1 with surgical procedure), a partial obliteration of vascular malformation was obtained. One patient died due to main cerebral hemorrhage onset. Fifty-five patents had a good outcome.

Over the years, there has been a progressive shift in the treatment of intracranial dural AVFs with endovascular treatment considered the first line of therapy in the majority of cases. However, until recently, e-dAVFs were considered almost exclusively surgical lesions because of the danger of inadvertent embolization of the ophthalmic artery, inability to consistently achieve a distal microcatheter position, and the relative ease of the surgical exposure and ligation of these lesions. Endovascular techniques continues to evolve, and in many cases it is possible nowadays to achieve microcatheter position very close to the actual point of fistualization through either a transarterial approach or retrograde transvenous route. These advances along with improvement in the embolic agents available have made endovascular therapy both safer and more effective in selected patients with e-dAVFs. A more aggressive stance toward endovascular therapy in recent years is reflected in our series where 46 % of treated patients underwent successful endovascular therapy. Based on this observation, we recommend consideration for endovascular treatment in patients at higher surgical risk (for example elderly with multiple medical comorbidities) and when distal catheterization very close to the point of fistualization can be safely achieved through a transarterial or a retrograde transvenous approach.

In conclusion, our series confirms previous reports of a higher male predominance in patients with e-dAVFs. It also shows that, as observed in the case of other "aggressive" dAVF locations, more and more lesions are diagnosed in patients with symptoms other than hemorrhage or in asymptomatic patients. In a modern series, selected patients can be effectively and safely treated through an endovascular route, although the majority of patients continue to benefit from open surgical ligation.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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