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Rupture of ethmoidal dural arteriovenous fistula – indication for emergency surgery (clinical cases)

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Objective of this paper is to demonstrate the variants of surgical treatment of ethmoidal dural arteriovenous fistulas (eDAVFs), propose a treatment algorithm for ruptured and unruptured eDAVFs taking into account the literature data. This article presents 4 clinical cases of patients with eDAVFs. The arterial supply of eDAVF was performed by the ethmoidal branches of the ophthalmic artery in all cases. There were 3 ruptured eDAVF with formation of intracranial hematoma and one unruptured eDAVF. Three patients underwent open surgical intervention, one – transvenous eDAVF embolization. Complete exclusion of eDAVF without complications in the postoperative period was confirmed in 3 patients, 1 patient died because of consequences of hemorrhage.

Patients with eDAVF rupture required immediate surgical intervention due to extremely high risk of repeat rupture. The main techniques of eDAVF treatment are open surgical intervention and endovascular embolization. Patients with unruptured eDAVF should be referred to Federal neurosurgical centers for endovascular treatment.

Keywords: ethmoidal dural arteriovenous fistula, arteriovenous fistula of the anterior cranial fossa, transarterial embolization, transvenous embolization

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INTRODUCTION

Ethmoidal dural arteriovenous fistulas (eDAVF) are a rare vascular anomaly, accounting for approximately 10 % of all intracranial dural arteriovenous fistulas. eDAVF s are located in the anterior cranial fossa at the level of the ethmoid plate, supplied with blood by the anterior ethmoidal branches of the ophthalmic arteries, and much less frequently by the branches of the middle meningeal artery, with the following drainage into the ascending cortical vein of the frontal lobe and then into the superior sagittal sinus, and less frequently into the olfactory vein. Drainage is usually unilateral. Due to the absence of “damping” dural sinuses at the location of eDAVF, this anomaly belongs to type 3 according to the Borden classification or types 3 and 4 according to the Cognard classification. The risk of life-threatening rupture of the eDAVF is considered extremely high [1–5], which, regardless of the presence of clinical symptoms, dictates an active surgical approach.

Despite the fact that various surgical methods of treating eDAVF are described in the literature, there is no consensus

on the algorithm of surgical tactics for this vascular anomaly.

The purpose of the paper is to demonstrate the options for surgical treatment of eDAVF and propose a possible algorithm for treatment of ruptured and unruptured eDAVF taking into account the literature data.

The article presents our clinical cases and highlights issues of the rare occurrence of this pathology with a fairly pathognomonic localization of hemorrhage, active surgical tactics for ruptured eDAVF and the possibilities of endovascular treatment.

CLINICAL CASE 1

Male patient K., 64 years old. It is known from the anamnesis that after awakening the patient developed speech disorders. The patient was admitted by an ambulance team to the city clinical hospital, where, during the examination, a intracerebral hemorrhage in the left frontal lobe was revealed due to the computed tomography (CT) of the brain. According to the brain CT angiography eDAVF was detected in the left frontal lobe (Fig. 1).

The patient refused from surgical treatment. After discharge, he began to notice an increasing headache, that is why he has been consulted by a neurosurgeon at the Federal Center of Brain Research and Neurotechnologies of the Federal Medical and Biological Agency (FCBRN of FMBA of Russia). The headache and partial motor aphasia were noted in the neurological status during examination. Due to the high risks of repeated rupture of the eDAVF, the patient was urgently admitted to the neurosurgical department for surgical treatment (in 26 days from the episode of primary hemorrhage).

Given the presence of ruptured eDAVF and intracerebral hematoma, a decision was made to perform open surgery. A bifrontal approach was used to reach the basal regions of the left frontal lobe (Fig. 2).

There was an eDAVF supplied by the anterior ethmoid artery of the left ophthalmic artery in the left-sided area of the ethmoid bone plate. The clipping of the anterior ethmoid artery and removal of the intracerebral hematoma in the left frontal lobe were performed.

The postoperative period was uneventful, the patient was discharged in 9 days after surgery. The postoperative examination data are presented in Fig. 3.

CLINICAL CASE 2

Male patient P., 67 years old. At night 17.10.2021 a sharp headache and slight weakness in the right leg were suddenly developed. The patient performed magnetic resonance imaging of the brain, which revealed a hemorrhage in the left frontal lobe, and he was urgently admitted to the neurosurgical department of the city clinical hospital. CT of the brain and brain CT angiography showed an intracerebral hematoma in the left frontal lobe with a volume of 10.8 cm³ and eDAVF was diagnosed (Fig. 4).

The patient was urgently moved to the FCBRN of FMBA of Russia for surgical treatment. Minimally invasive transfrontosinus approach, clipping of the anterior ethmoid artery and removal of the intracerebral hematoma of the left frontal lobe (on 4th day after the episode of primary hemorrhage) were performed. The postoperative period was

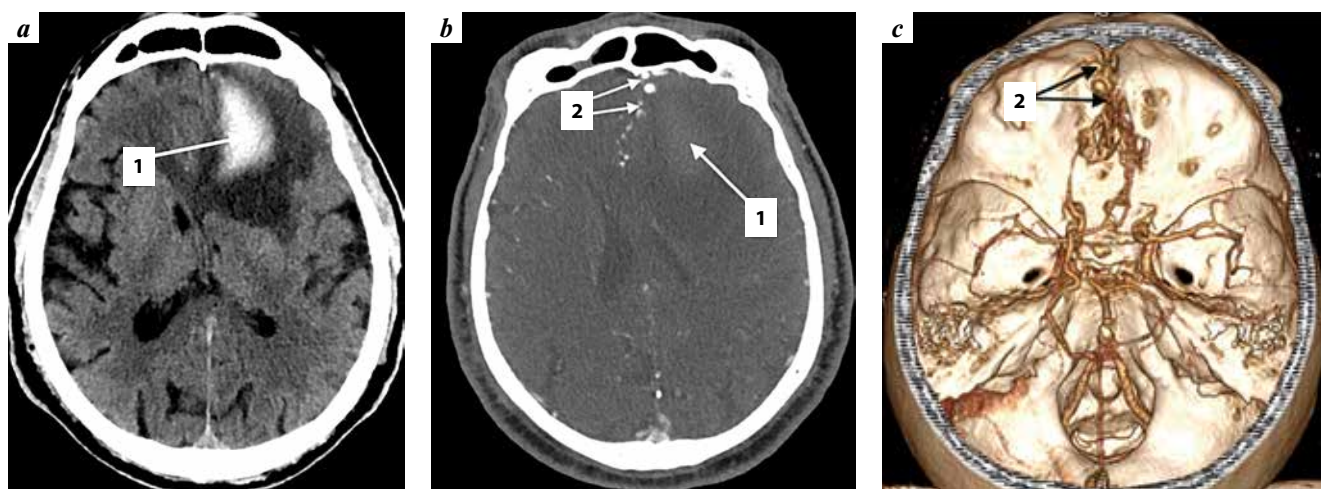


Fig. 1. Computed tomography (CT) of the brain and CT angiography of the intracranial cerebral arteries of patient K, 64 years old: a – Brain CT, axial projection; b – multiplanar reconstruction (MPR); c – 3D shaded surface display (SSD). 1 – subacute intracranial hematoma of the left frontal lobe; 2 – ethmoidal dural arteriovenous fistula

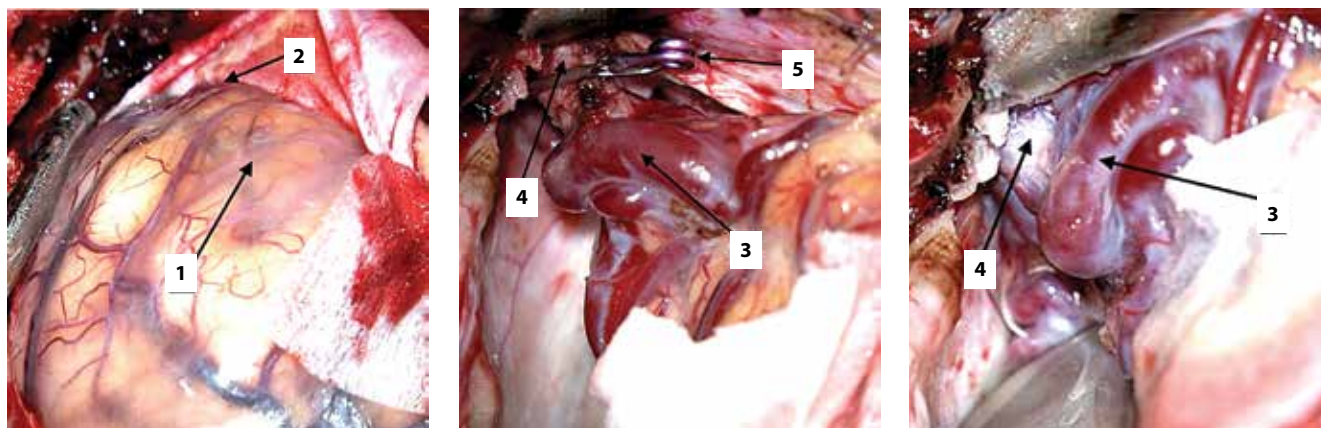


Fig. 2. Intraoperative images of patient K., 64 years old. 1 – left frontal lobe; 2 – longitudinal fissure; 3 – emissary vein, 4 – anterior ethmoidal artery, 5 – clipping of the anterior ethmoidal artery

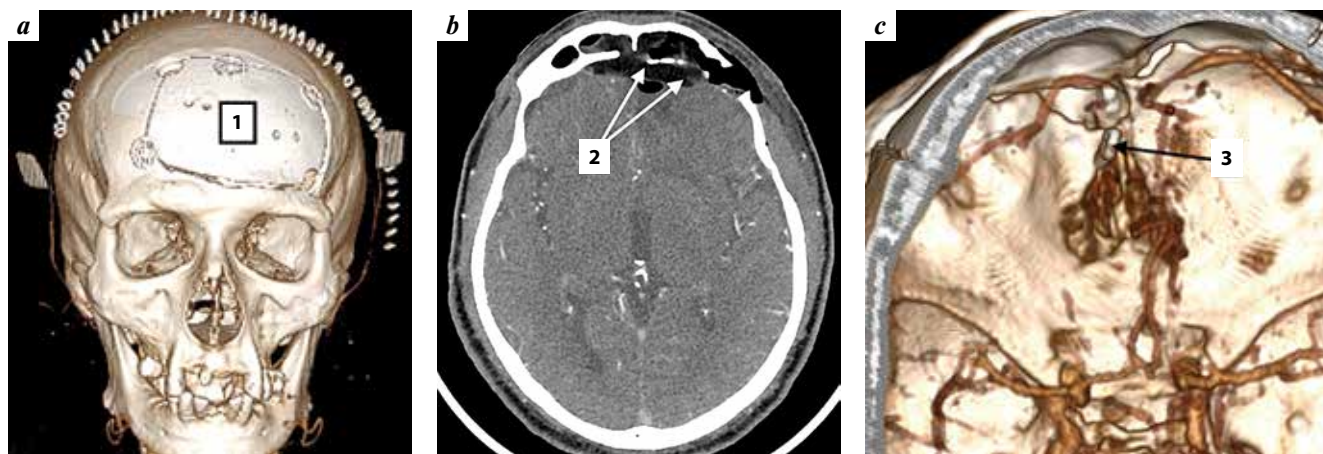


Fig. 3. Computed tomography (CT) of the brain and CT angiography of the intracranial cerebral arteries of patient K., 64 years old. Postoperative control: a – 3D reconstruction (SSD); b – multiplanar reconstruction (MPR); c – 3D shaded surface display (SSD). 1 – trephination area; 2 – surgery area; 3 – clip

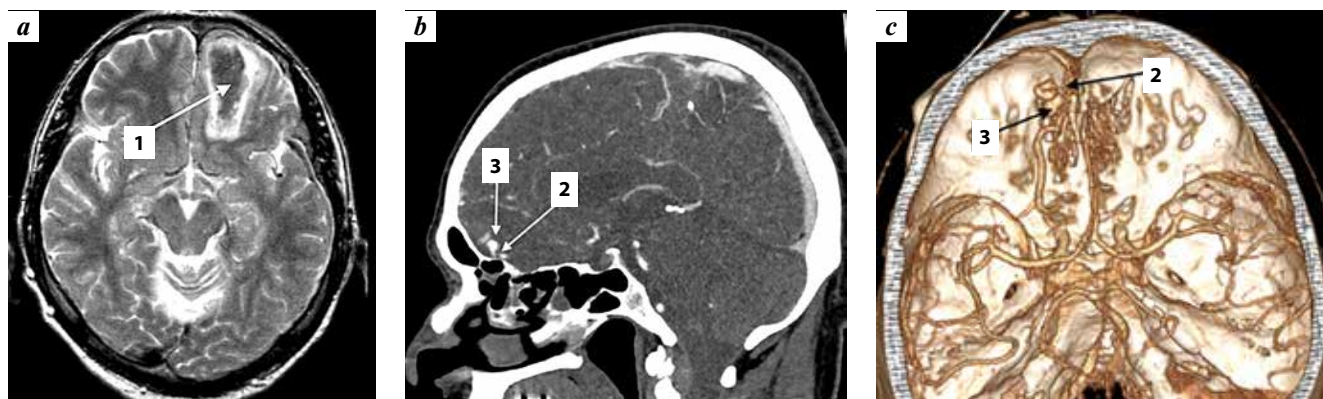


Fig. 4. Computed tomography (CT) of the brain and CT angiography of the intracranial cerebral arteries of patient R., 67 years old: a – brain CT, axial projection; b – multiplanar reconstruction (MPR); c – 3D shaded surface display (SSD). 1 – intracranial hematoma of the left frontal lobe; 2 – anterior ethmoidal artery; 3 – ethmoidal dural arteriovenous fistula



Fig. 5. Computed tomography (CT) of the brain and CT angiography of the intracranial cerebral arteries of patient R., 67 years old. Postoperative control: a – 3D reconstruction; b – multiplanar reconstruction (MPR); c – 3D shaded surface display (SSD). 1 – trephination area; 2 – surgery area; 3 – clip

uneventful, the patient was discharged after 7 days. The postoperative examination data are presented in fig. 5.

It should be noted that in both presented cases (clinical cases 1 and 2) the hematoma had a very pathognomonic location for ruptured eDAVFs – the anterior basal medial part of the frontal lobe.

CLINICAL CASE 3

Male patient Y., 58 years old was admitted to the city clinical hospital by an ambulance team. The neurological status at admission was the following: level of consciousness – sopor (10 points on the Glasgow Coma Scale), pupils $D = S$, smoothing of the left nasolabial fold, left-sided hemiplegia,

tendon reflexes $D > S$, Babinski's sign on the left. During examination, according to brain CT and CT angiography of the cerebral intracranial arteries, parenchymatous-ventricular hemorrhage (the volume of the parenchymatous part of the hematoma is 40 cm^3) and eDAVF on the right were revealed (Fig. 6).

On an emergency basis (1 hour after the hemorrhage episode), the decompressive craniotomy was performed in the right frontal-parietal-temporal region. During removal of the intracerebral hematoma of the right frontal and parietal lobes, a repeated rupture of the fistula with intense arterial bleeding occurred; clipping of the varicose veins with multiple clips was performed in the absence of clear visualization of the bleeding source. The hematoma from the right frontal lobe was completely removed. A ventricular drain was installed in the anterior horn of the right lateral ventricle (Fig. 7, a).

The patient was in the intensive care unit in the postoperative period, brain CT was performed in 12 hours after

the operation. (Fig. 7, b). There was no evidence of recurrent hemorrhage. The patient died on the 8th day due to cerebral edema and multiple organ failure.

This clinical case shows the difficulties during management of massive intraoperative bleeding in emergent surgery. Clipping and/or coagulation of the draining veins often do not stop the bleeding and, as a rule, it is necessary to look for a hypertrophied anterior ethmoid artery (eDAVF).

CLINICAL CASE 4

Male patient C., 43 years old. The patient felt a sharp headache and nausea on 26.02.2019, against the physical activity. He was admitted to the primary vascular department, brain CT revealed the subarachnoid hemorrhage, saccular aneurysm of the anterior communicating artery (ACoA), multiple miliary aneurysms of both middle cerebral arteries and eDAVF without rupture. The endovascular subtotal

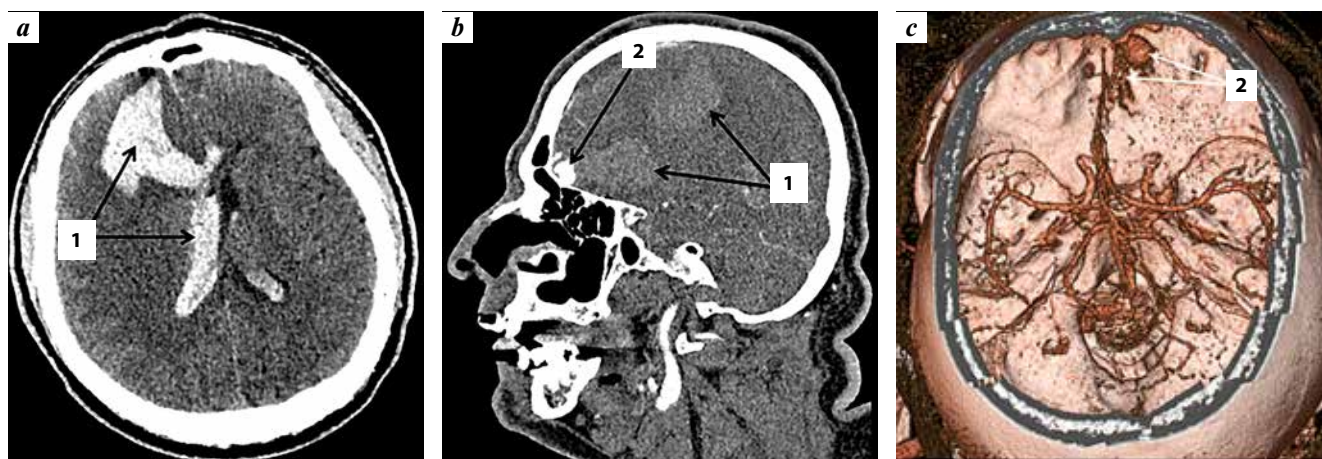


Fig. 6. Computed tomography (CT) of the brain and cerebral CT angiography of patient U., 58 years old: a – brain CT, axial projection; b – multiplanar reconstruction (MPR); c – 3D shaded surface display (SSD). 1 – parenchymal intraventricular hemorrhage; 2 – ethmoidal dural arteriovenous fistula

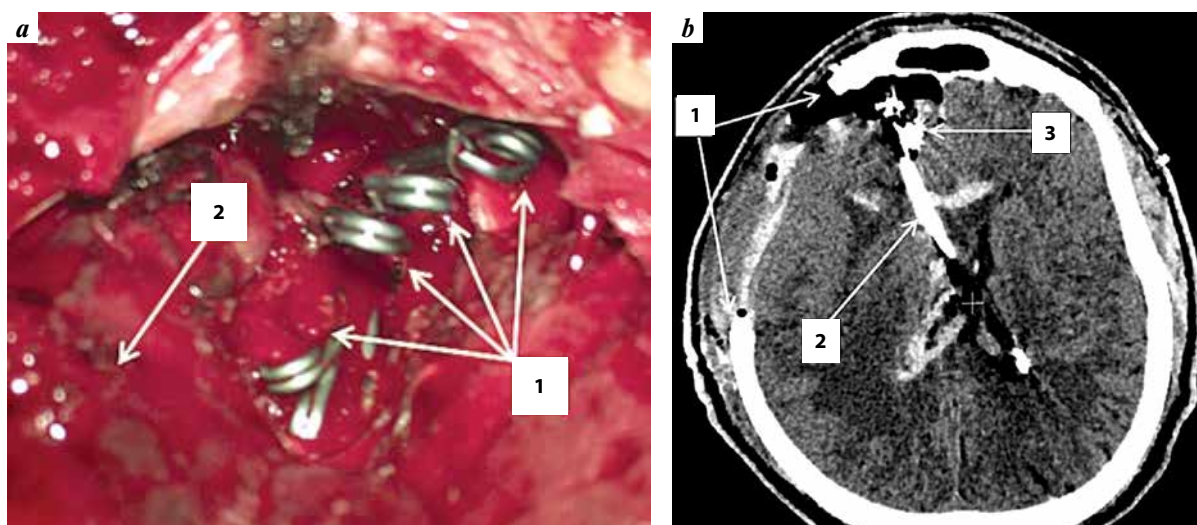


Fig. 7. Data of patient U., 58 years old: a – intraoperative image: clipping of varicose veins with vascular clips in the absence of clear visualization of the source of bleeding. 1 – clips, 2 – frontal lobe; b – brain CT, axial projection, postoperative control. 1 – area of surgical intervention; 2 – ventricular drain; 3 – clips

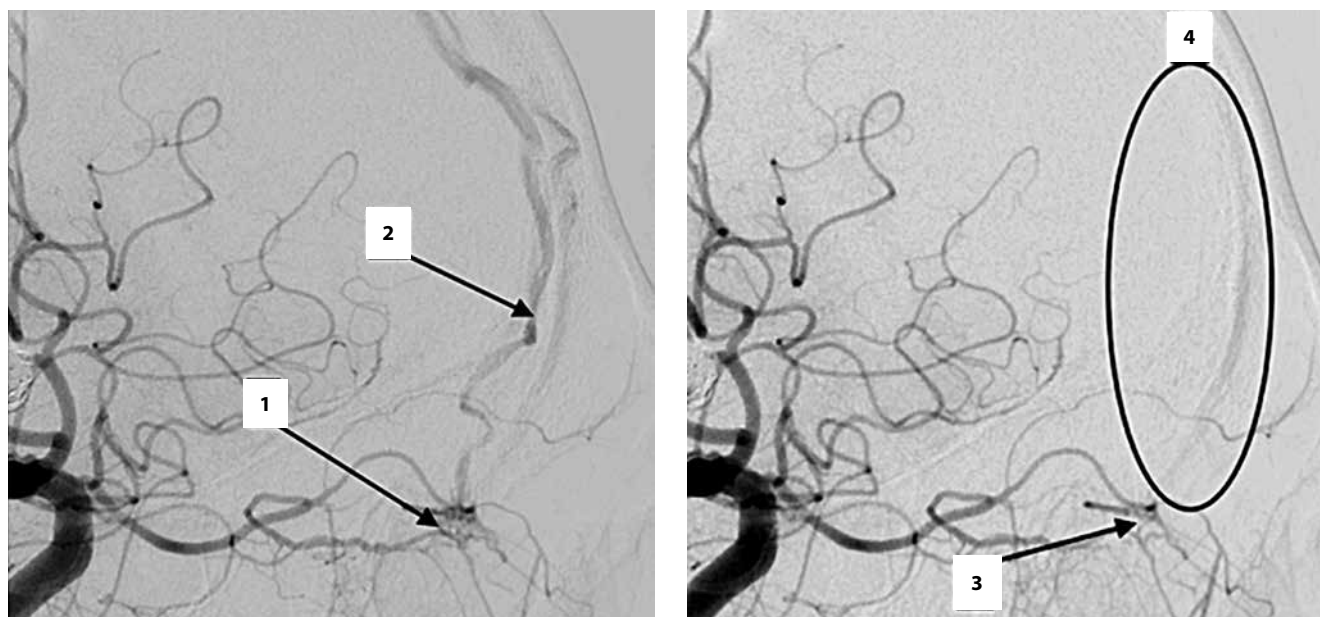


Fig. 8. Cerebral angiography of patient S., 43 years old. 1 – ethmoidal dural arteriovenous fistula; 2 – ascending cortical vein; 3, 4 – anterior cranial fossa dural arteriovenous fistula in the area of crista galli, totally embolized, no signs of contrast agent filling the fistula

embolization of the anterior communicating artery aneurysm with microcoils was performed as the 1st step of treatment. The postoperative period was uneventful.

The control angiography in 1.5 year revealed the recanalization of the ACoA aneurysm, miliary aneurysms of both middle cerebral arteries and eDAVF (without dynamics). The endovascular treatment of the ACoA saccular aneurysm with stent assistance was performed followed by the total transvenous embolization of eDAVF using non-adhesive composition Squid12 (Fig. 8).

The postoperative period was uneventful, the patient was discharged on the 4th day after the operation.

DISCUSSION

Ethmoidal dural arteriovenous fistulas are considered an “aggressive” vascular disease due to the high risk of hemorrhage, which is caused by the anatomical features of the fistula structure – direct drainage of arterial blood into the cortical veins bypassing the microcirculatory net and dural sinuses. In 59 % of cases, the draining vein has focal varicose veins, which indicates the hemodynamic overload of the fistula [3, 6, 7].

S. Giannopoulos et al. (2019) indicate that the risk of bleeding in this vascular anomaly reaches 91 % [8]. According to H. Duffau (1994) and J. M. van Dijk (2004), repeated hemorrhages occur in 35 % of cases within 2 weeks after the primary hemorrhage and are accompanied by more severe consequences than the first hemorrhage. The annual rate of neurological complications is 15 %, and the annual mortality is 10.4 % [9, 10].

The routine method of treating eDAVF is microsurgical disconnection of eDAVF by clipping the anterior ethmoid artery. According to worldwide literature, the success rate

of microsurgical treatment is very high and ranges from 85 to 100 % [11–14]. However, surgical intervention is associated with some risks, the frequency of complications ranges from 0 to 20 %. Complications include cerebrospinal fluid leakage, meningitis, damage of the frontal lobe and olfactory nerve [11, 12, 14–16].

Endovascular fistula disconnection can be performed via transarterial or transvenous approaches. Transarterial embolization of eDAVF using various adhesive compositions is a well-known treatment method; the occlusion rate varies from 12.5 to 63.6 % [11, 12, 17]. However, embolization via the ophthalmic artery is considered technically challenging and dangerous due to possible occlusion of the central retinal artery, which can develop in the case of excessive glue reflux, migration of glue fragments during the catheter removal as well as dissection or vascular spasm. T. Robert and J.M. Abrahams described embolization of eDAVF via the middle meningeal artery. This approach may be used as an alternative to embolization via the ophthalmic artery; however, blood supply of eDAVF via the middle meningeal artery is rare [15, 18].

Transvenous embolization of eDAVF is a rare intervention [8, 15, 19–22]. One of the main problems with the transvenous approach is the difficulty of the catheterization of the draining vein, which is often tortuous. According to the literature data, the frequency of occlusions during transvenous embolization is high, but all patients in the described cases were carefully selected according to the criterion of «availability» of venous drainage, so these results should not be summarized [8, 15, 19, 20, 22].

We suppose that the choice in favor of one or another treatment method should be made in each clinical case individually, and the risks and benefits of each procedure

should be taken into account and discussed with the patient. Indeed the choice of treatment method will mainly depend on the anatomical features of the fistula structure, as well as on the experience of the operating surgeon. The angiographic architecture of each eDAVF plays an important role in the prognosis of the disease, especially if the patient has a complex pathology of the cortical veins, when the risk of bleeding with any manipulation is extremely high. Careful preoperative determination of the anatomy and individual features of the venous drainage of the eDAVF

is crucial for the optimal choice of treatment and achieving a good surgical outcome.

CONCLUSION

The main method of choice for the treatment of ruptured eDAVF is the open surgery, and unruptured ones are treated with endovascular embolization. Patients suffered from ruptured eDAVF require the immediate surgical intervention due to the extremely high risk of rerupture.

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Authors' contributions

I.V. Senko: obtaining data for analysis, analyzing the data obtained, developing of research design, scientific editing of the article;
E.S. Ryzhkova: development of research design, review of publications on the topic of the article, article writing;
S.A. Melchenko, I.V. Grigoriev, D.V. Khovrin: obtaining data for analysis, scientific editing of the article.

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