

## Case Reports &amp; Case Series

## Successful endovascular treatment of extra-intracranial arteriovenous malformation using a combination of liquid non-adhesive embolic agents

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## ABSTRACT

Congenital malformations of blood vessels, including arteriovenous malformations (AVMs), are a less common pathology, which remains one of the most diagnostically and therapeutically complex diseases for treatment. Patients with vascular abnormalities often receive an erroneous diagnosis and inadequate treatment. Depending on the abnormality, such improper treatment can lead to potential long-term functional and cosmetic consequences. Existing treatment options include surgical resection, endovascular embolization, and a combination of these methods. To date, there is no pharmacotherapy available that allows you to radically treat this pathology. Despite recent advances, AVMs are rarely cured and may require multi-stage therapy throughout life. The article presents a clinical case of successful endovascular treatment of extra-intracranial arteriovenous malformation of the frontoparietal region in a young girl with a more pronounced extracranial component of AVM on the front and frontoparietal region with cosmetic and functionally significant defect. We performed double-stage embolization of AVMs with a combination of non-adhesive liquid embolic agents with high and low viscosity. We got good angiographic, clinical and cosmetic treatment results.

### 1. Introduction

Arteriovenous malformations are rare congenital vascular lesions that occur in any area of the human body. They can be life threatening due to potential massive hemorrhage. Based on the characteristics of the endothelium, Mulliken and Glowacki [1] (1982) classified vascular lesions into 1) hemangiomas – vascular tumors and 2) vascular malformations. AVM of scalp are rare occurrences among vascular lesions. Various names being used to describe the vascular malformations of the scalp include aneurysm cirroid, aneurysma serpentinum, aneurysm racemosum, plexiform angioma, arteriovenous fistula and arteriovenous malformation [2]. Both categories of vascular lesions have different etiologies and clinical features. Hemangiomas are vascular tumors that

show endothelial hyperplasia, which increases due to rapid cell proliferation. They are usually absent at birth, but develop during the 1st year of life and then manifest.

Vascular malformations are congenital structural malformations with a normal metabolic rate of endothelial cells, which are present at birth, but usually become noticeable at a later age. A rapid increase in malformations is usually caused by trauma or hormonal changes during puberty or pregnancy. An increase in these lesions is associated with a change in pressure and flow, expansion of the vascular channels, bypass and collateral proliferation, and not cell proliferation. Moreover, based on the characteristics of blood flow, vascular malformations can be divided into low level lesions and high - level lesions. Low lesions include capillary, lymphatic and venous malformations, while high

**Abbreviations:** AVM, arteriovenous malformation; CT, computed tomography; SCAG, selective cerebral angiography; MRI, magnetic resonance imaging; ECA, external carotid artery; ICA, internal carotid artery.

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lesions include arterial and AVM [3].

Hemangiomas are formed due to lack of differentiation in the early stages of embryogenesis, however, vascular malformations arise due to disturbances in the late stages of angiogenesis, which leads to the preservation of embryonic arteriovenous anastomosis. [4] The shunt from high pressure to low pressure in the compartment may have a ripple effect. A shunt vein expands and thickens, and an increase in flow velocity explains the expansion of arteries [5].

AVMs are rare, they account for only 1.5% of all vascular abnormalities, and 50% of lesions are located in the oral and maxillofacial regions [6].

We presented a clinical case of successful endovascular embolization of extra – intracranial arteriovenous malformation with a more pronounced extracranial AVM component of the frontoparietal region in a young patient with a cosmetic and functional defect due to the expansion of the superficial temporal and frontal (supratrochlear) veins. We carried out a double-stage total embolization of AVMs with a combination of non-adhesive liquid embolic agents with high and low viscosity with a good angiographic, clinical and cosmetic effects.

## 2. Clinical case

Patient S., female, 30 years old

Complaints of noise in the head, headaches, periodic dizziness, a cosmetic defect of the frontoparietal region due to the expansion of the saphenous vessels on the forehead, temples and scalp.

Anamnesis: since childhood, there has been a swelling of blood vessels under the skin on the forehead and scalp, dizziness, noise in the head, headaches. Notes an increase in volume and prevalence after childbirth.

According to computed tomography (CT) with head vessels contrasting, extra - intracranial arteriovenous malformation (AVM) of the frontoparietal region, with a more pronounced extracranial component, was revealed (Fig. 1).

Visually, the patient has a cosmetically significant defect in the frontal region of the face, in view of the marked expansion of the superficial temporal and frontal (supratrochlear) veins (Fig. 2). The

synchronous pulsation was determined.

Diagnosis: Anomaly brachiocephalic and cerebral arteries. Extra – intracranial arteriovenous malformation of the frontoparietal region.

In order to determine further treatment tactics, the patient underwent selective cerebral angiography (SCAG): According to the SCAG: extra - intracranial arteriovenous malformation of the frontoparietal region with afferents from the right and left superficial temporal artery (Fig. 3), facial artery, anterior meningeal arteries was revealed (Fig. 4).

Given the nature of the lesion, angioarchitectonics, blood supply, and drainage of the AVM, a decision was made on staged endovascular embolization.

## 3. Operative technique

The first step was the embolization of afferents from the left superficial temporal artery.

Under general anesthesia, the right femoral artery was punctured. An angiographic catheter 5F on a guidewire 0,035" was inserted through the inserted introducer 6F. Catheterized left external carotid artery (ECA) and was performed cerebral angiography. The angiographic catheter has been replaced with the guiding catheter Guider Softip™ XF (Boston Scientific) 6 French (F). Installed in the ECA. DMSO – compatible flow-dependent braided microcatheter Sonic® (BALT) 1.5F with a tear-off portion of 1.5 cm, is inserted into the distal part of the left superficial temporal artery.

At the first stage, we performed a Sonic® (BALT) 1.5 F microcatheter in an AVM, embolized the fistula part of the AVM with three doses of the non-adhesive liquid embolic agent SQUID™ 18 (BALT) (Fig. 3). And racemose part and dural AVM perforants were embolized with a non-adhesive liquid embolic agent with a very low viscosity PHIL™ 25% (MicroVention). On control angiography embolized AVM is determined (Fig. 5).

The second stage, after 3 months, the embolization of afferents from the right superficial temporal artery was performed according to the same strategy as for the first time (Fig. 6).

The early postoperative period was uneventful. Access point without features. The pulse on the femoral artery was preserved, no hematomas

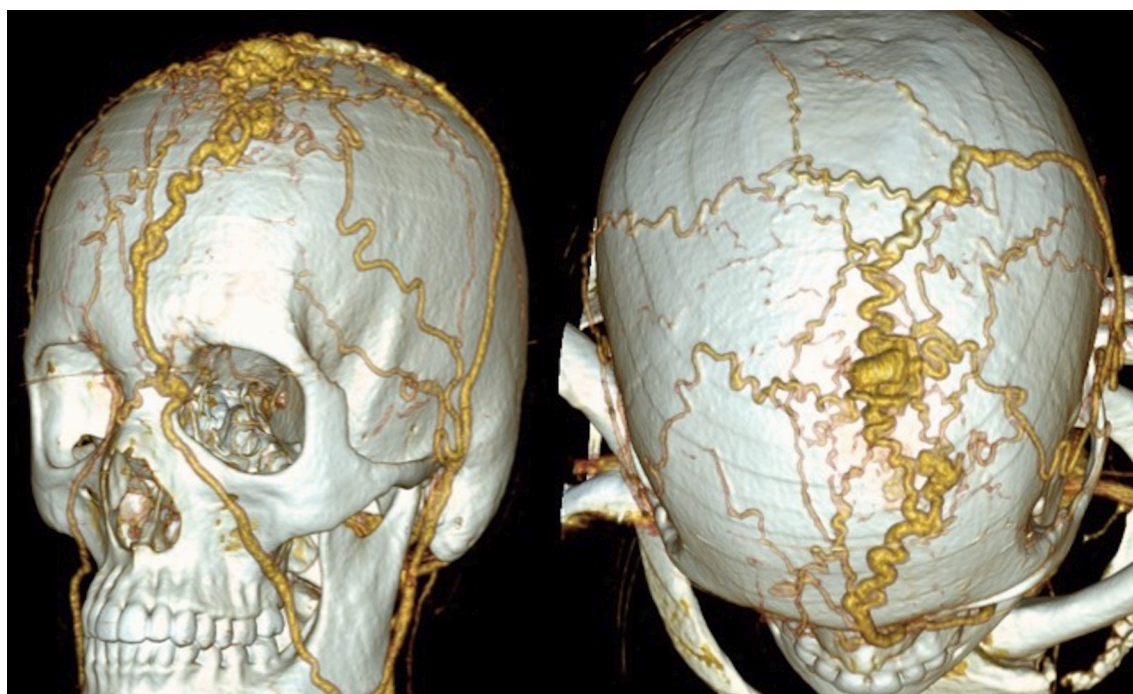
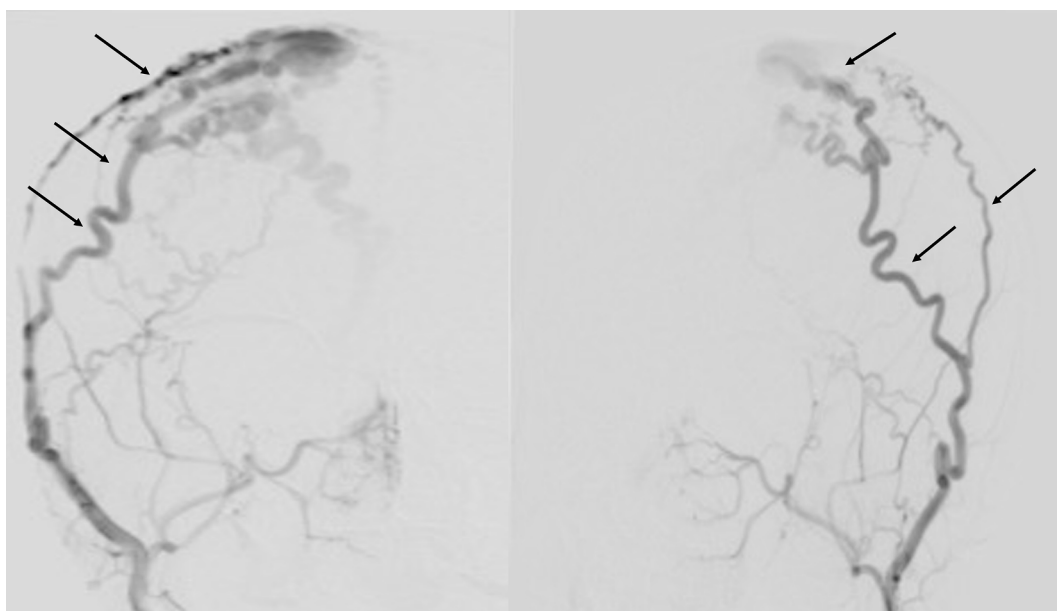


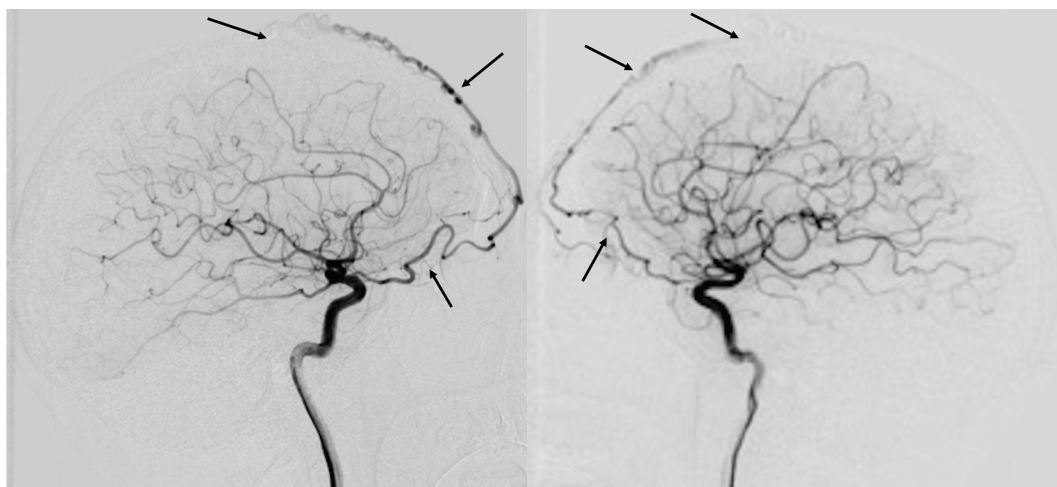
Fig. 1. CT – angiography of the vessels of the head, 3D – reconstruction: Arteriovenous malformation of the frontoparietal region with afferents from the right and left superficial temporal artery, facial artery.



**Fig. 2.** The appearance of the patient before surgery. The expanded subcutaneous vessels of the frontal area are determined.

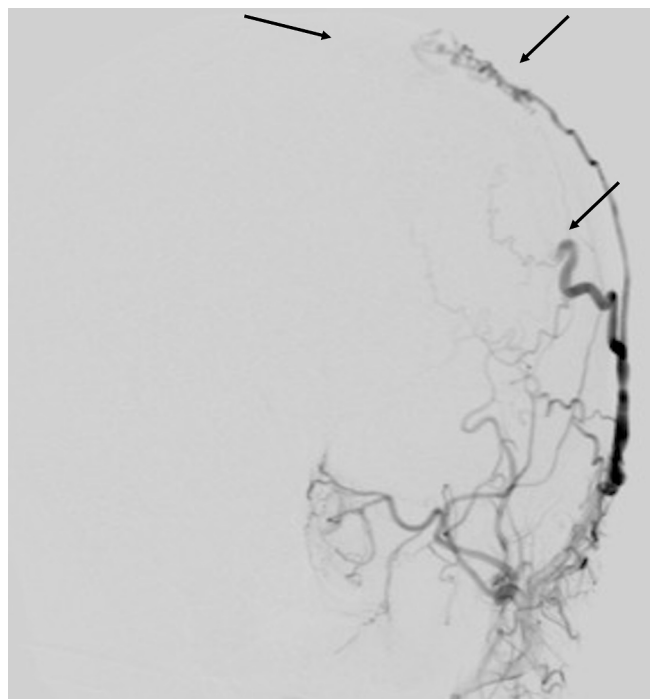


**Fig. 3.** Cerebral angiography of the external carotid arteries (ECA), direct and lateral projection: Arteriovenous malformation of the frontoparietal. Afferents of the superficial temporal arteries on the right and left.



**Fig. 4.** Cerebral angiography, right and left side projections, angiography of the internal carotid arteries (ICA). Afferents from the left and right anterior meningeal artery.





**Fig. 5.** Cerebral angiography, direct projection, angiography of the left ECA. Embolized section of the AVM.



**Fig. 6.** Cerebral angiography, direct projection, angiography of the right ECA. Embolized section of the AVM.

were detected. Neurological status of the patient without features. Discharged in satisfactory condition on the 3rd day after the intervention.

In the distant period, three months after the intervention, a significant decrease in the dilated subcutaneous vessels of the frontal and parietal regions is determined. Satisfactory cosmetic and clinical effects of the operation are noted (Fig. 7).

#### 4. Results and discussion

AVMs are vascular abnormalities consisting of abnormal anastomoses between the arterial and venous system, which causes blood bypass. AVMs are the most aggressive form of vascular abnormality, which can lead to significant deformation and functional impairment. Rapid vascular malformations usually occur in childhood and puberty [7]. Kohout et al. their study found that AVMs were present at birth in 59% of cases, in children in 10% of cases, in adolescence in 10% of cases, and in adulthood in 21% of cases [8].

Various diagnostic tools are available for diagnosing vascular lesions, such as color Doppler ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), and magnetic resonance angiography. Type of malformation can be confirmed by ultrasound using color Doppler studies. CT with iodinated contrast identifies AVM as a highly effective diagnostic method and can demonstrate soft tissue involvement as well as dilated vessels [9]. Digital subtraction angiography is the gold standard investigation for these lesions, which provides a road map of arterial supply and venous drainage [10].

AVM treatment is difficult due to the involvement of normal tissue in the vasculature with a high relapse rate [11]. The treatment of this pathology mainly consists of surgery, vascular embolization, or a combination thereof [12]. The goal of embolization is to block the vessels that feed the AVM. Various materials can be used for embolization, such as polyvinyl alcohol particles, muscles, gel foam, cyanoacrylate, metal spirals and collagen [4].

In this case, the patient underwent a two-stage embolization. This tactic was not chosen by chance. The patient had an extensive high-speed AVM with a pronounced fistulous part from the pool of superficial temporal arteries and facial arteries. Given the localization and involvement of a large area of the scalp and facial part in the AVM, we decided to perform a two-stage procedure with an interval of three months in order to prevent necrotic complications from the soft tissues of the head and face.

In this case, we used a combination of two non-adhesive liquid embolic agents of high and very low viscosity. For the fistulous component of AVM, embolization was performed with an embolic agent SQUID™ 18 with a high degree of viscosity, for the racemose component we used an embolic agent with a very low viscosity PHIL™ 25%. According to our observations, this combination is optimal for embolization of extensive mixed AVMs.

#### 5. Conclusion

This clinical case has shown the efficacy and safety of the use of endovascular techniques in the treatment of extra – intracranial AVMs.

The stages of the procedure and the use of various non-adhesive liquid embolic agents can prevent and minimize complications associated with necrotic lesions of the soft tissues of the face, eliminate paresis of the facial nerves, and form an adequate reduction in blood flow after primary embolization. It also allows achieving good angiographic, clinical and cosmetic treatment results in patients with this pathology.

Unfortunately, frequent relapses of the disease indicate the need for constant monitoring of patients.

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#### 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.



Fig. 7. Appearance of the patient 3 months after surgery. No «tattoo» - effect and dilated saphenous vessels.

#### Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.inat.2020.101008>.

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