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# Intraoperative Ultrasound-Guided Resection for Extracranial Arteriovenous Malformations of the Head and Neck

Giacomo Colletti[a], Riccardo Nocini[b], Linda Rozell-Shannon[c], Luigi Chiarini[a], Alexandre Anesi[a], Margherita Dessy[d]

**Abstract**

**Objectives:** Arteriovenous malformations (AVM) are a highly aggressive and difficult to manage disease. Treatments such as proximal ligation, partial embolization, and incomplete surgical removal often lead to a relapse that is worse than the "virgin" condition. Thus, when surgery is chosen with curative purposes, a radical resection must be obtained. Color Doppler can offer a way to more reliably plan resection margins and, if needed, extend the excision of the AVM to obtain radicality.

**Methods:** Nine patients with S1–S3 facial AVMs underwent radical surgical resection with the aid of Color Doppler Ultrasound (CDUS). CDUS was used to define the margins of the exeresis, to check for residual disease and, in case, to extend the resection. Primary closure or local flaps were used to reconstruct the defect.

**Results:** Histology showed AVM-free margins in the 5 patients where CDUS showed no signs of residual disease. In 4 cases, the resection was secondarily widened according to the US findings. Here, the histology showed remnants of the nidus in the part close to the original resection and no disease elsewhere in the remainder of the specimen. At a 24–62 months follow-up, no patient showed signs of relapse.

**Conclusion:** The use of CDUS seems to guarantee a higher degree of cure rate for "healable" AVMs that are managed by means of surgery. Since its introduction in our clinical practice, we routinely use this method in the management of AVMs that are amenable to radical removal.

**Keywords:** angioma, hemangioma, arteriovenous, AVM, radical, surgery, ultrasound, US, Doppler

## Introduction

Arteriovenous malformations (AVMs) are a rare congenital vascular anomaly believed to be caused by a genetic mutation in the endothelial cells of the capillary plexus during the early stages of embryogenesis.[1,2] An extracranial arteriovenous malformation (EAVM) is an AVM that occurs outside of the brain (extracranial). This report focuses on the EAVM.

Both EAVMs and brain AVMs are characterized by a tangle of tortuous vessels with a direct shunt between the arterial and venous systems. This tangled cluster is referred to as the "nidus."

In the natural history of EAVMs, there is the tendency to increase in size and invade adjacent tissues, frequently in response to trauma or hormonal changes. For example, a surgical trauma, such as the incomplete resection of the EAVM, may induce an aggressive recurrence with potential irreversible consequences to eradicating the disease. The same holds true for partial or proximal embolization. Also, hormonal surges, such as those related to pregnancy, can stimulate rapid growth of the EAVM. These exacerbations of the disease, along with a high risk of relapse after treatment, point out an analogy between EAVMs and tumors. Therefore, as with the treatment of neoplasms, radical excision with free margins on the anatomic specimens is one of the most relevant issues as far as long-term prognosis is concerned. In cervico-facial EAVMs, planning a radical surgical resection gains a critical relevance since it must be balanced between the risk of relapse and the predictable impairment of function and/or esthetics.

A Color Doppler Ultrasound (CDUS) device has effectively been used to provide a visual representation of the movement of blood through blood vessels with the help of ultrasonic waves. The use of CDUS has been described in previous papers[3–6] as a useful method to diagnose and monitor treatment and management of EAVMs. Additionally, the use of intraoperative ultrasound (IUS) is widely adopted in

[a]uniMORE, Modena, Italy; [b]University of Verona, Department of Otolaryngology, Verona, Italy; [c]VBF, Latham, New York; and [d]University of Sassari, Sassari, Italy

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Correspondence: Giacomo Colletti, MD, Piazza della Repubblica 21, 20124 Milan, Italy (giacomo.colletti@unimore.it).



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the management of central nervous system AVMs.[7–9] Few papers discuss the role of IUS to assess resection margins of spinal EAVMs.[10] CDUS has also been used in conjunction with neuronavigation to help in the resection of brain AVMs.[11] Finally, CDUS has been used to aid in the removal of uterine EAVMs.[12] To our knowledge, the use of CDUS has not been described before in the intraoperative management of EAVMs of the head and neck.

The purpose of this article is to illustrate a novel technique for the intraoperative guidance and assessment of the resection margins of cervico-facial EAVMs using an ultrasound Color Doppler transducer.

## Materials and methods

Being this a retrospective analysis, an Institutional Review Board exemption was granted.

We enrolled 9 patients that were referred to our department presenting with cervico-facial EAVMS from 2016 to 2021. Each patient was studied with a contrast enhanced Magnetic Resonance Imaging, an angio-Computed Tomography, and a preoperative CDUS evaluation. The latter was performed by the same surgeon (G.C.) who also did all the surgeries (Table 1).

All EAVMs were staged as per SECg Staging System developed by Colletti et al.[13] Inclusion criteria were:

- Presence of a S1 or S2 AVMs regardless of E, C, or g
- Presence of a S3 AVM which was locally complicated (C2) or growing (g+)

Exclusion criteria were:

- Presence of an S3 AVM with little symptoms (C0, C1) and not growing (g–)
- Presence of an S4 AVM (incurable AVM)
- Presence of an AVM involving deeper tissues not amenable to allow for US assessment

Mean age at the time of surgery was 27 years (range 19–37). Anatomical regions involved included 3 upper lips (1 pericommissural and 2 of the philtrum), 1 lower lip, 1 infraorbitary-cheek region, 1 glabellar region, 1 ear while 2 AVMs were in the temporal region.

## Technique

Two of the 9 patients referenced in this study have been managed by means of a combined endovascular-surgical treatment plan and 7 patients have been managed with surgery only.

On studying each lesion, when the EAVM was deemed resectable without any significant bleeding, or when the EAVM was too peripheric to be addressed with endovascular treatment, then surgery, alone, was chosen. Conversely, when the size of the malformation required a pre-surgical reduction of vascularity, and was amenable, then preoperative embolization was performed. The combined approach was chosen for 1 temporal malformation and 1 infraorbital-cheek EAVM. Embolization was performed 24 hours before surgery by the interventional radiologist, with patients in mild sedation with intravenous administration of propofol under anesthesiologic monitoring. Through a femoral artery approach, a superselective embolization of the malformation was obtained using Ethylene Vinyl Alcohol Copolymer (ONYX™, Medtronic, Santa Rosa, CA, USA) until most feeders of the EAVM were obliterated, to obtain a surgical field with as little bleeding as possible. Surgery was performed under general anesthesia with the recommendation not to use any curare during the whole surgical procedure. In this way, the facial nerve could be monitored and all its branches near the malformation detected, to protect them. A sterile field was prepared leaving the entire hemiface drape-free.

In the first patient, we planned a conventional "clinically guided" resection (Figures 1–3). After this, we performed a CDUS check with waveform analysis (with a 4–12 MHz linear transducer) and we were able to detect suspicious images beyond the margins of resection (Figures 4–6). Given this information, we extended the resection until we could see that all the field was free from disease in terms of US images. We sent the extended margins separately for histological examination (Figure 7). The defect was closed with local flaps (Figure 8). At a 58-month follow-up, the patient is free from relapse (Figure 9).

In all 8 subsequent cases, we planned the resection by identifying the margins of the EAVM with CDUS and spectral analysis to distinguish normal vessels. The surgical margins were then marked on the skin (Figures 10 and 11). The EAVM was then excised accordingly, paying attention not to damage any adjacent facial nerve branches. However, if some are encountered during the procedure, they should be isolated and spared unless there is an evident embedding of the branch in the pathologic tissue.

After resection, the linear 4–12 MHz transducer is covered with sterile drapes and the excision margins and the deep wound bed are again checked to investigate any residual disease. Any areas of turbulent and pulsatile blood flow found during the Special Doppler analysis, with the typical characteristics of EAVMs (high velocities, low resistance, spectral broadening in the arteries, and pulsatile flow in the venous component) are noted, then the resection is widened in the remaining area. This procedure is repeated

**Table 1.**

**Patients Demographics and AVM Features**

| Patient | Age | Sex | Site | Stage |
|---|---|---|---|---|
| D.R. | 29 | F | Upper lip | S3C2+ |
| T.D. | 33 | M | Infraorbital | S2C1– |
| M.P. | 37 | M | Upper lip | S3C2+ |
| C.A. | 26 | F | Glabella | S2C1+ |
| S.A. | 28 | M | Temporal | S3C1+ |
| E.E. | 25 | M | Ear | S3C2+ |
| V.F. | 19 | M | Upper lip | S3C2+ |
| S.S. | 23 | F | Lower lip | S3C2+ |
| L.V. | 27 | M | Temporal | S2C1+ |

Abbreviation: AVM, arteriovenous malformation.

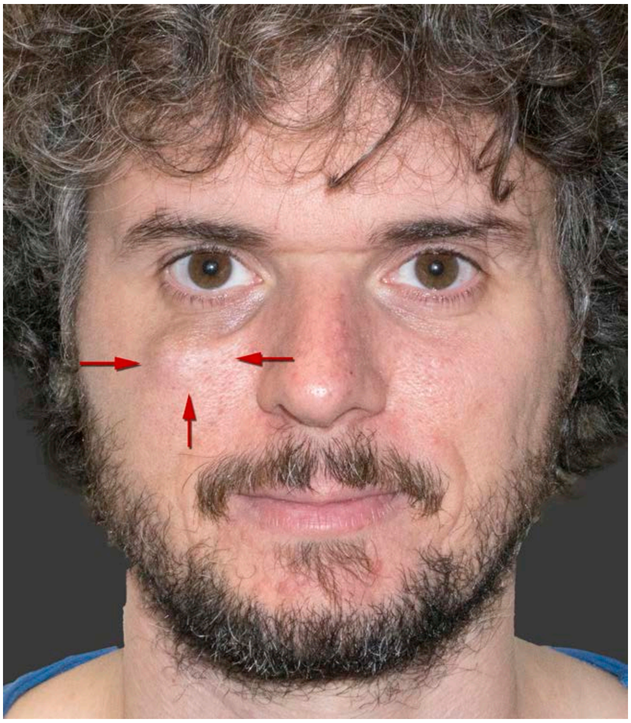

**Figure 1.** Patient 1 with AVM of the right zygoma. The limits of the disease are ill defined if just the clinical features are used (arrows). AVM indicates arteriovenous malformation.

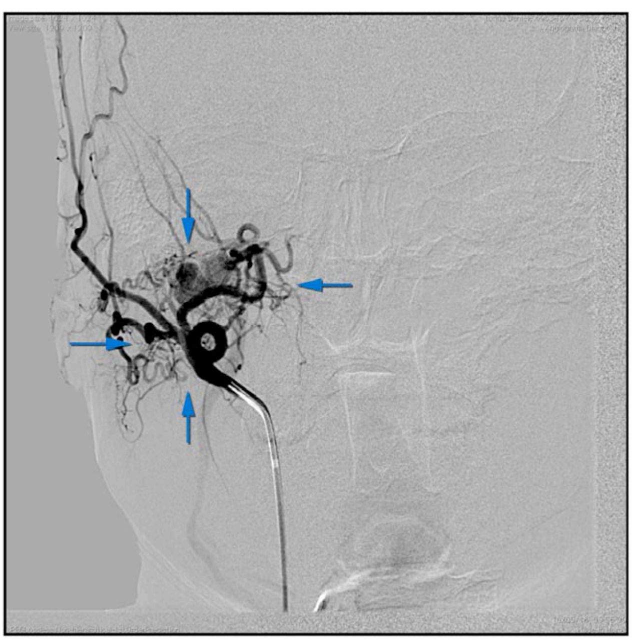

**Figure 2.** Patient 1. Angiogram shows an AVM of the right zygoma (arrows). AVM indicates arteriovenous malformation.

until healthy margins at the cutaneous and deep borders are reached (Figures 12–15).

Pathological and genetic examination was performed in all the tissue specimens excised (Figure 16).

The residual defect was closed by means of direct closure or local flaps (Figure 17).

No relapse was detectable at a 62-month follow-up (Figure 18).

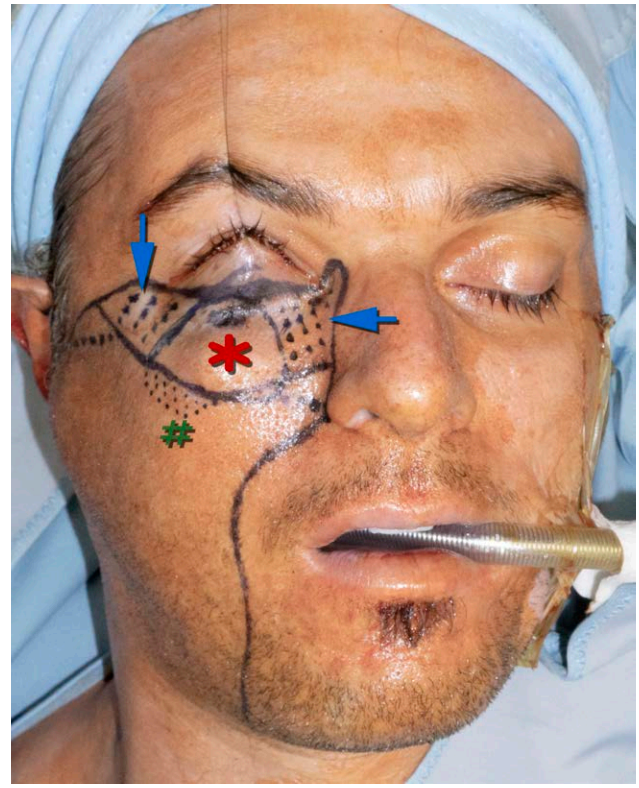

**Figure 3.** Patient 1. Planning of resection. Clinically planned excision (asterisk); extension of full thickness resection (arrows); subcutaneous resection (number sign).

## Results

Four of the 9 patients included in this study required a further extension of the primary resection, that was planned preoperatively, after checking the residual margins. In all 9, histopathological examination reported the presence of the typical vascular pattern of EAVMs in every principal specimen and in every secondary resection made by the support of US Doppler. Interestingly, no signs of residual EAVM were found in the resection margins. Follow-up was made by a US examination every 2–3 months and a contrast enhanced Magnetic Resonance Imaging every 6 months in the first year after the operation, followed by periodic checks settled on the clinical and radiological findings in each patient.

The mean follow-up is 29 months (range 24–62 months) and all the patients are EAVM disease free as of November 2021.

## Discussion

As previously stated, many EAVMs may resemble neoplasms for their propensity to grow and infiltrate the surrounding tissues spontaneously or in response to traumas and/or hormonal changes. In particular debulking or incomplete resection, or embolization as the sole treatment, may result in an abrupt expansion induced by hypoxia and the subsequent release of Vascular Endothelial Growth Factor and other proangiogenic factors (hypoxia-induced factor) by the cells inside the remaining nidus. The result is a condition that is often much worse and much more aggressive than the "virgin" AVM.

Surgical excision is considered the most effective means in the treatment of EAVMs[14]; thus, radicality and free resection margins should be considered essential concepts for curative treatment.

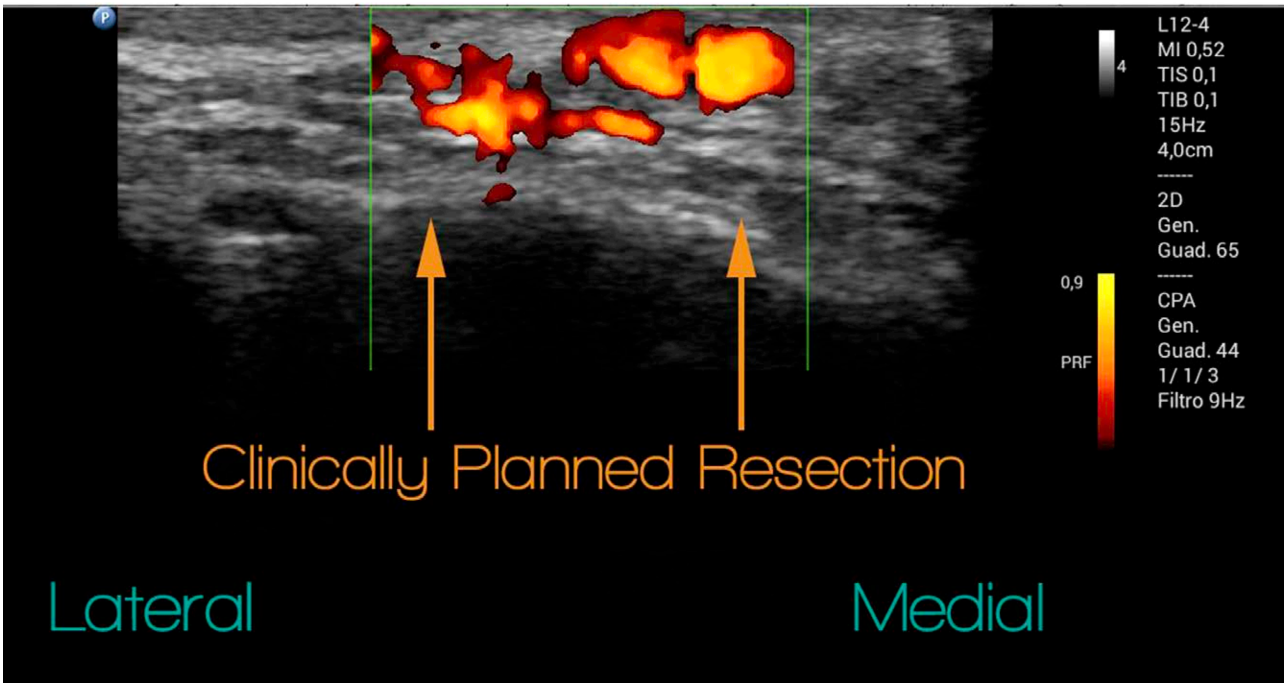


**Figure 4.** CDUS of the clinically planned resection. The AVM appears to involve the margins (orange arrows and green lines). AVM indicates arteriovenous malformation; CDUS, Color Doppler Ultrasound.

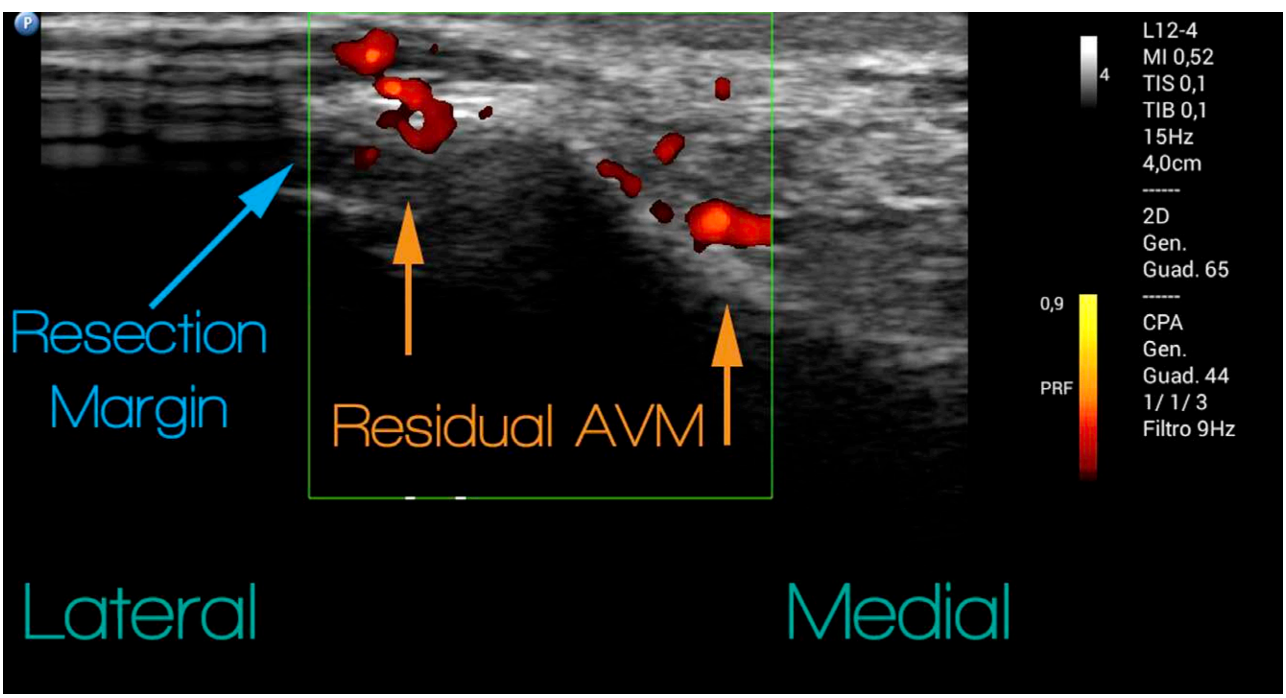


**Figure 5.** CDUS of the soft tissues medial to the resection. Residual disease is detected and the excision extended accordingly. CDUS indicates Color Doppler Ultrasound.

In head and neck EAVMs, the clinical and radiological margins of the EAVM may not be clear. Therefore, the surgeon may underestimate the margins, leading to a potential relapse, or overestimate them, leading to an overtreatment and all the related consequences in terms of aesthetic and functional impairment.

Once surgical planning is made, based on clinical and radiological preoperative imaging only, and the resection is performed, there is no other way than an angiography to test the radicality of surgery. Some authors have proposed this to control the margins of the EAVM.[15]

The aim of this research was to find a safe and effective means capable of providing adjunctive guarantees to reach clean surgical margins in EAVM resection, thus enhancing cure rate. Color Doppler and Spectral Doppler US were utilized effectively and are a painless, noninvasive, and cost-effective technique for assessing these lesions. Remarkably, it proved a reliable means to prevent relapses

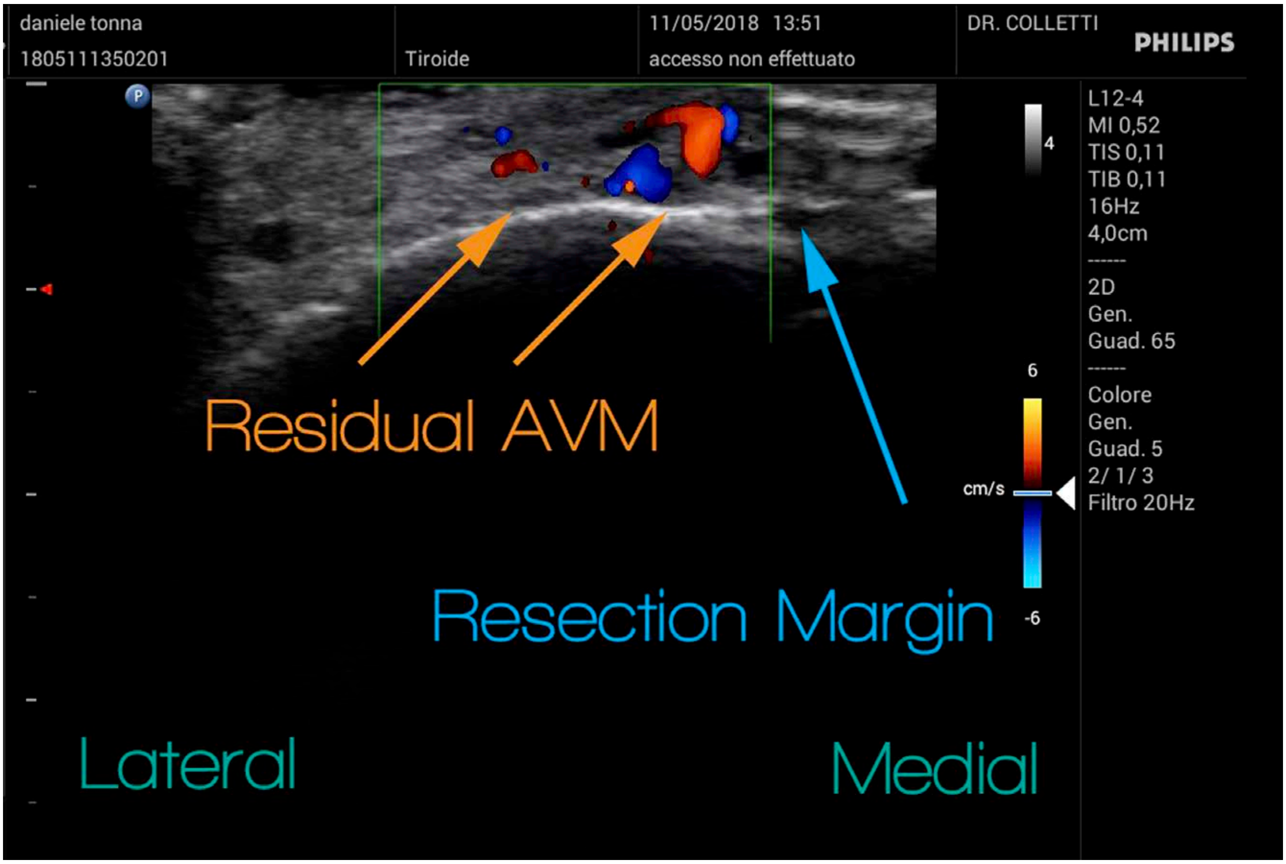


**Figure 6.** CDUS of the soft tissues lateral to the resection. AVM is still present and the exeresis extended. AVM indicates arteriovenous malformation; CDUS, Color Doppler Ultrasound.

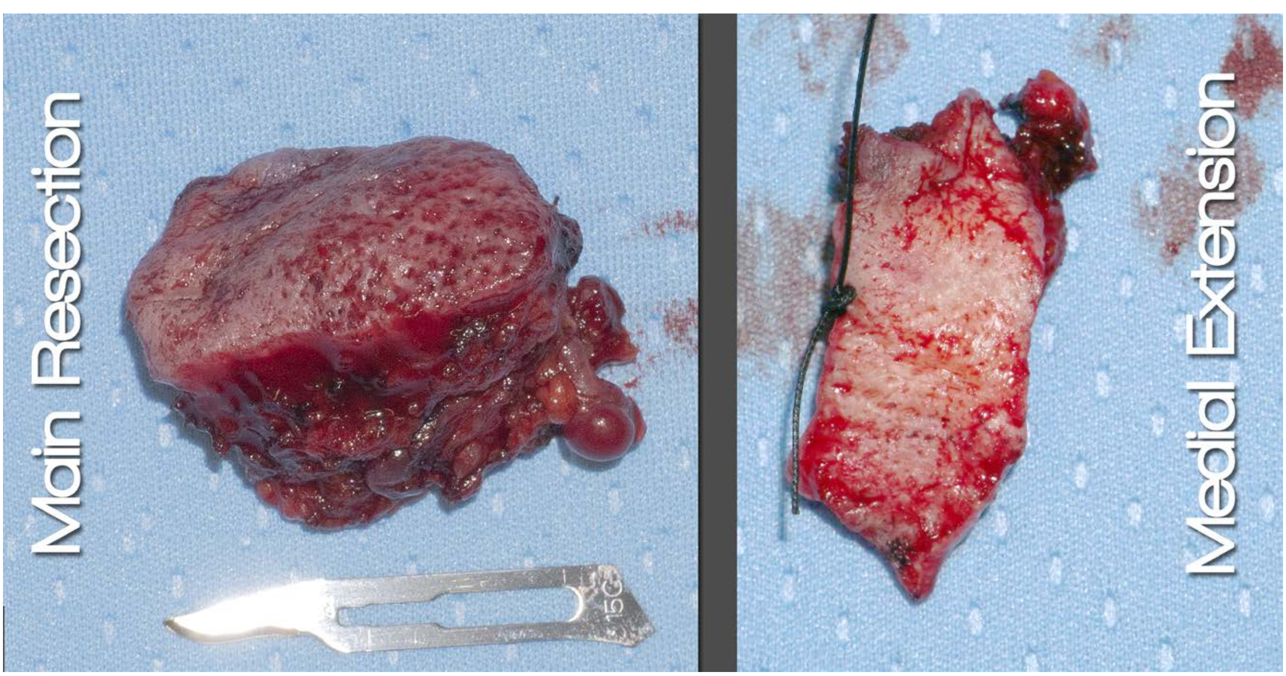


**Figure 7.** Specimen of the main resection and the medial extension.

as revealed at 24 months minimum follow-up (longest follow-up 62 months).

The first intraoperative use of ultrasound (a 1-dimensional finger probe with no Color Doppler) for the surgical management of EAVMs was described in 1979 in 2 different papers. In these papers, however, a Doppler probe was used to localize an intestinal EAVM and to determine the proximal and distal points at which abnormal signals were present,

**Figure 8.** Patient 1. Closure of the defect with local flap.

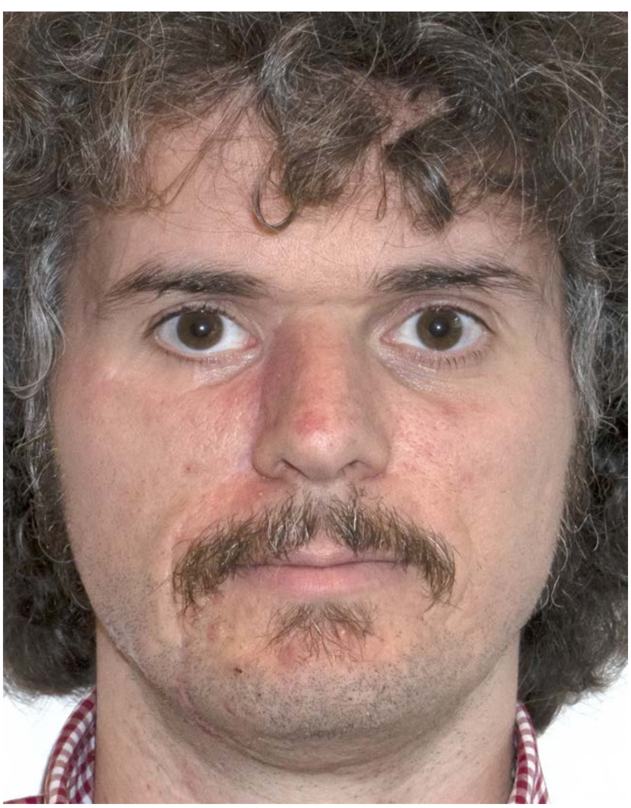

**Figure 9.** Patient 1: clinical appearance at 58 months after surgery.

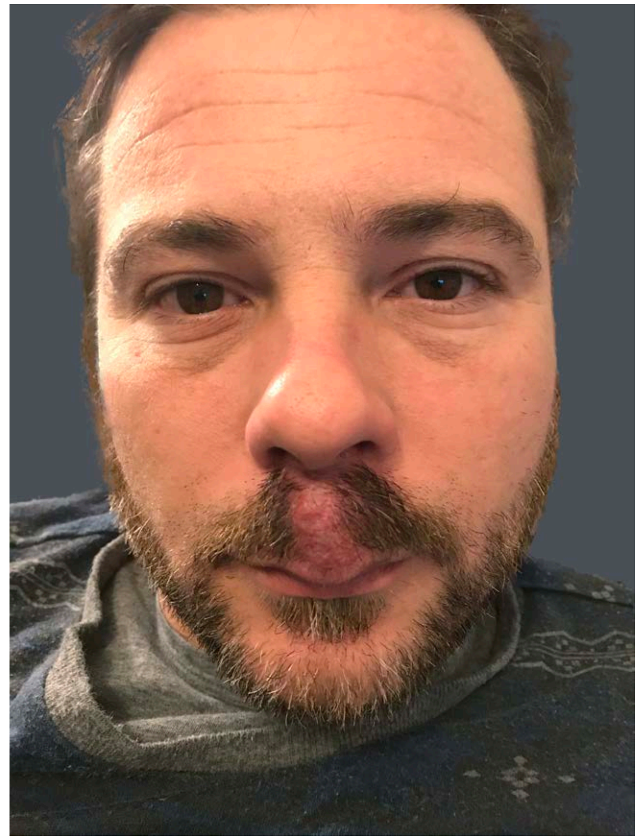

**Figure 10.** Patient 2 with an AVM of the philtrum. AVM indicates arteriovenous malformation.

**Figure 11.** Patient 2: the margins of the lesion are drawn relying on the CDUS. CDUS indicates Color Doppler Ultrasound.

to guide the surgical resection.[16,17] Afterward, in the late 1980s, different ultrasound techniques were reintroduced in the intraoperative management of EAVMs in neurosurgery, where it was used for intraoperative diagnosis and evaluation of resection margins in spinal cord EAVMs.[18,19] Subsequently, CDUS,[7] Spectral Doppler,[20] and CEUS[21] were introduced as a guide for complete resection of intracranial AVMs.


**Figure 12.** Patient 2: after completion of the resection, a new CDUS reveals residual AVM (arrow). AVM indicates arteriovenous malformation; CDUS, Color Doppler Ultrasound.

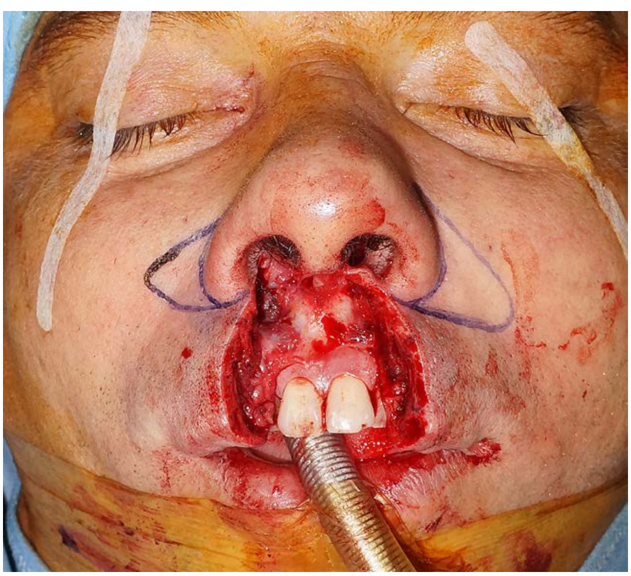

**Figure 13.** Patient 2. the resection is extended until a silent CDUS is obtained. CDUS indicates Color Doppler Ultrasound.

Besides its use in neurosurgery, US has been occasionally described in the intraoperative management of pulmonary[22] and uterine[12,23] EAVMs.

To our knowledge, the utilization of intraoperative US was described in head and neck EAVMs only in one paper reporting a case of EAVM of the scalp. In this case, the Doppler US was used to guide thrombin injection and transarterial coil embolization of the malformation.[24] However, we discourage the sole embolization of EAVMs (and especially the use of coils) because it may produce a hypoxic and inflammatory environment that could likely result in neo-angiogenesis and worsening of the initial disease.[25]

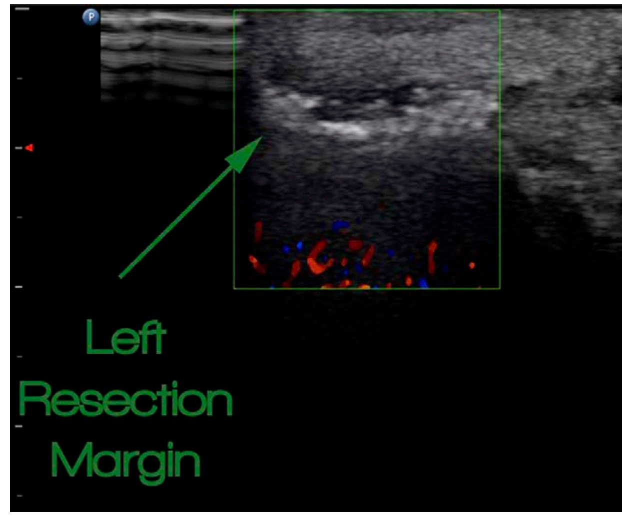


**Figure 14.** CDUS of the soft tissues beyond the left margin. CDUS indicates Color Doppler Ultrasound.

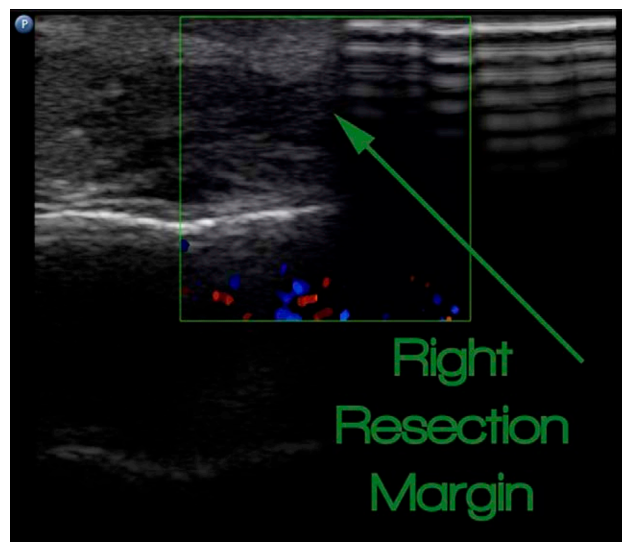


**Figure 15.** CDUS of the soft tissues beyond the right resection margin. CDUS indicates Color Doppler Ultrasound.

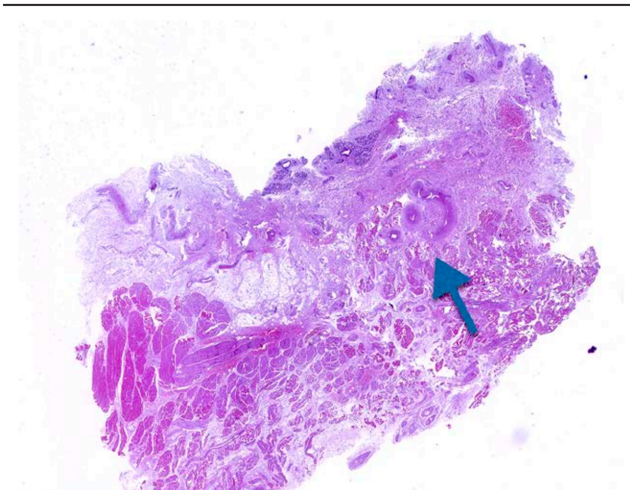

**Figure 16.** Patient 2: histology of the extended excision. Focal AVM nidus is revealed (arrow). AVM indicates arteriovenous malformation.

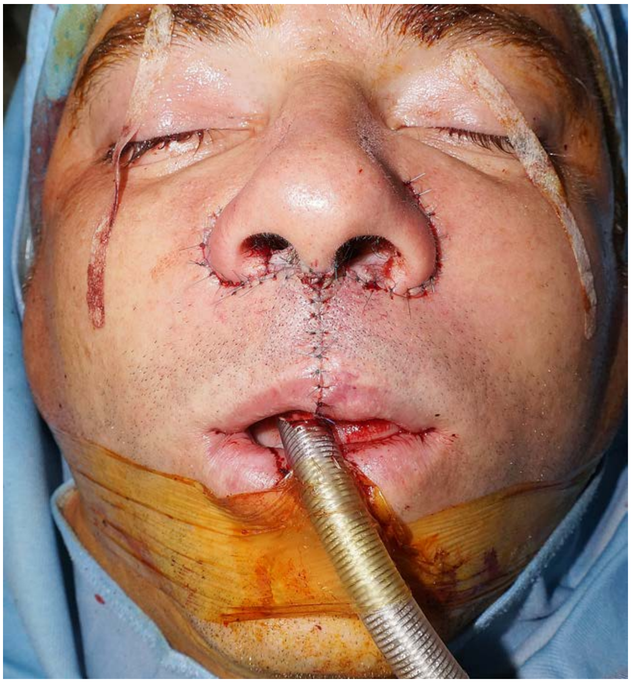

Figure 17. Patient 2: closure with local flaps.

Figure 18. Clinical appearance at 62 months follow-up.

## The basics of B-mode, Color Doppler, and spectral analysis US

In brightness mode US (B-mode), a linear transducer makes a scan of a tissue and the returned echo is converted in a 2D image and is visualized in a grey-scale. The higher the amplitude (ie, density of the tissue) of the reflected wave, the brighter the image represented. In this way, one can visualize and differentiate the structures, to evaluate their spatial relationships and their different densities. CDUS is based on the principle that the US wave emitted by the transducer, with a given frequency, is reflected by red blood cells moving within the vessel with a change of frequency directly proportional to the velocity of blood. The reflected wave is registered by the transducer and converted into a quantitative acoustic signal, giving the opportunity to measure the flow and vascularity.[4] CDUS collects information about the hemodynamics of the lesion. The blood flow approaching the transducer is displayed as a red color image, while blood flow moving away from the transducer is coded in a blue color, with a color brightness that is proportional to the flow velocity.[26,27] Pulsed wave Doppler permits the spectral analysis of the arterial and venous flow, showing the temporal trend of the Doppler signal, according to their amplitude and frequency, giving a representation of the distribution of flow velocity over time. Spectral analysis also enables measurement of the resistive index in the arterial vessels (RI = [peak systolic velocity − end diastolic velocity]/peak systolic velocity).

## Ultrasound features of EAVMs

In B-mode, the US features of the EAVM appear as a conglomerate of anechoic tortuous vessels. In addition, the surrounding, interspersed soft tissues may appear hyperechoic. The characteristic high flow of these lesions is revealed on the Color Doppler imaging. Additionally, the nidus is displayed as a mixture of signals in a tangled pattern, with high vascular density and multidirectional flow. Afferent and efferent draining vessels can also be outlined. Spectral Doppler analysis reveals high peak velocities (>20 cm/s), low resistance index (≤0.50),[6] and spectral broadening of the arterial waveform when compared to normal ones on the contralateral side (Figure 19). The venous flow velocity profile may show a replication of the systolic and diastolic phase (although with much lower velocity) usually found in arteries. This phenomenon is known as "arterialization" of the venous outflow and is typical of EAVMs and arterio-venous fistulas.[4,5,28–30]

These information are critical to identify the resection margins and any residual nidus on the remaining tissue and to differentiate between pathologic vessels and simple enlarged feeding arteries or draining veins.

In a recent paper, that was quite intriguing, the authors measured the value of wall shear stress (WSS) at different levels (common carotid artery, external carotid artery, and facial artery) of the principal feeding artery of superficial EAVM of the lower face and compared these values with the ones found in the respective arteries of a control group. Patients were studied with a MRA, an angiography, and a measure of WSS at time 0 and again, after 6 months. The authors found that the increase of WSS positively correlated with the progression of disease observed on the radiological and clinical examinations at the 6-month follow-up. Following these results, the measure of WSS would be able to discriminate between progressive and stable EAVM, thus being a reliable marker of progression, with the 92% of specificity, along with 58%, 92%, and 100% of sensibility at the measurement performed on the common carotid artery, external carotid artery, and facial artery, respectively. Thus, the artery more proximal to the nidus is the one wherein

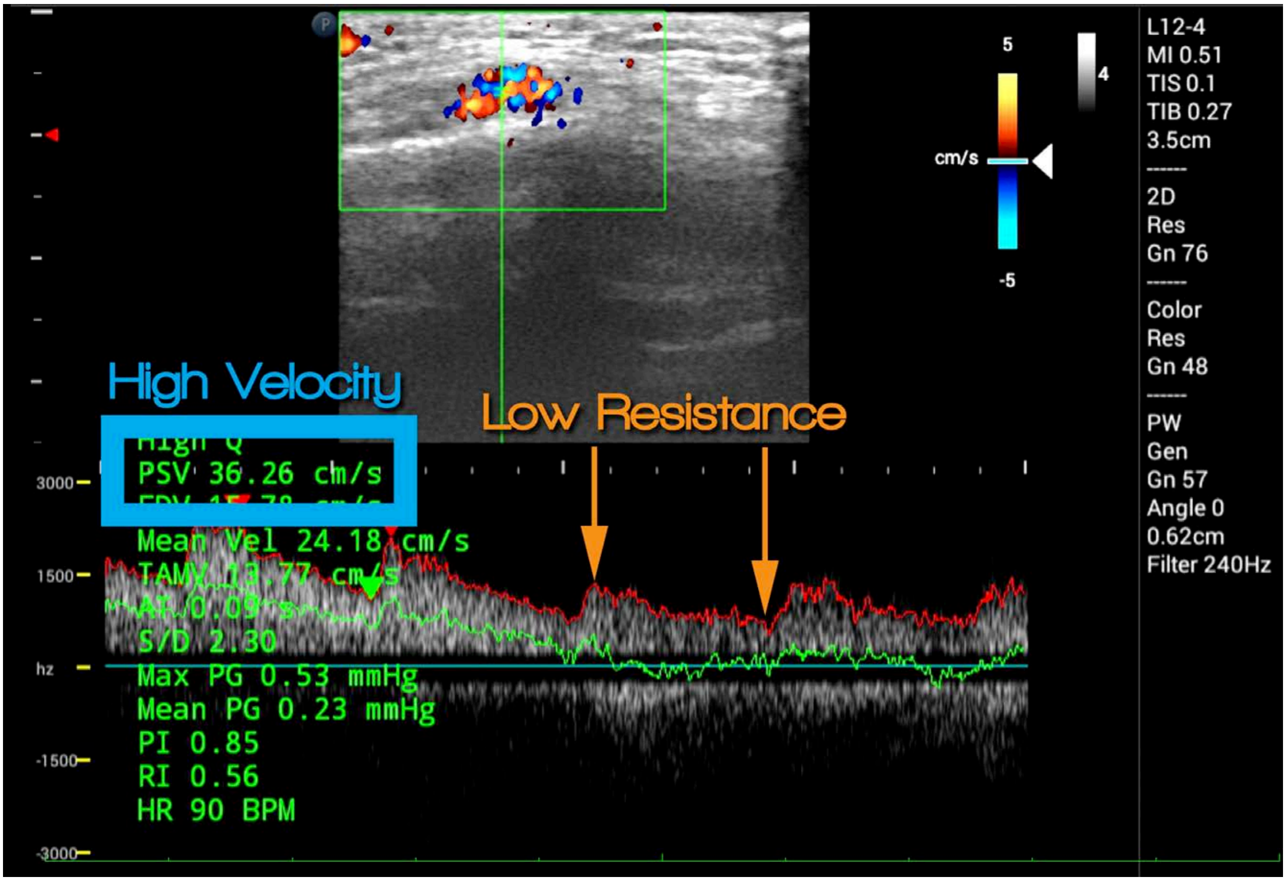


**Figure 19.** Typical power Doppler of AVM. The shape and absolute values show a high velocity flow (cyan square and arrow) and a low resistance shape (orange arrows). AVM indicates arteriovenous malformation.

it is registered the most significant variation of WSS.[31] The application of this method could represent a true valid measurement in the follow-up of EAVM and maybe become an invaluable means for the definition of treatment indication.

US and its different applications have multiple advantages in the diagnosis, management, and follow-up of EAVM. US is free from ionizing radiation, it does not need contrast medium injection, it is painless and noninvasive, can be rapid when performed by a trained operator, and is cheaper than other types of imaging. However, the main limits of the US are its inability to evaluate deep or bony components of malformations because of the inadequate penetration of sound wave into such tissues. Additionally, the operator-dependence of the technique, which is strictly connected to the expertise of the sonographer, can lead to potential false positive or negative results.

## Conclusion

US Doppler, according to this study, should be considered a reliable means in the diagnosis and follow-up of EAVMs.

Preliminary results in the intraoperative evaluation of surgical resection margins of EAVMs seems to be encouraging, providing a real time monitoring of the radicality of surgery thus giving the opportunity to enhance the cure rate as suggested by 0% rate of relapse after more than 2 years follow-up.

Further development of this technique, such as the introduction of the evaluation of WSS on the feeding arteries, in a routinely follow-up protocol, may also provide a more precise indication of the progression of the disease, as well as provide support for this proposed methodology for utilizing IOUS for the guided resection of an EAVM.

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