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Cutaneous necrosis following endovascular embolization by using ethylene-vinyl alcohol copolymer for arterio-venous malformations

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ABSTRACT

Arteriovenous malformations (AVMs) are intricate vascular anomalies marked by aberrant connections between dysplastic arteries and veins, often leading to a spectrum of complications. Treatment of AVMs consists of endovascular embolization, surgical resection or a combined approach. The endovascular embolization is an important and popular alternative treatment to reduce circulation of the nidus and prevent the development of collaterals and recurrence. However, local and systemic complications are quite common after repeated embolization, and surgical treatment is often required to manage such complications. This article presents a comprehensive analysis of a specific case, wherein a 32-year-old male patient with a gluteal AVM underwent ethylene vinyl alcohol copolymer (EVOH, Onyx®) embolization, subsequently developing large cutaneous necrosis. The core objective of this study was to search existing literature and provide insights into the management strategies employed in this unique scenario.

Key words: Arterio-venous malformation, AVM, cutaneous necrosis, endovascular embolization, ethylene vinyl alcohol copolymer, EVOH.

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Introduction

Arteriovenous malformations (AVMs) are rare congenital disorders characterized by direct vascular communication between pre-capillary arteries and post-capillary veins through dysplastic vessels network called nidus. Peripheral AVMs are located most commonly on the head and neck region and rarely on the lower limb and trunk. Depending on size, location, stage and invasion of peripheral AVMs, it may cause serious functional, aesthetic and life-threatening problems, including disfigurement, pain, ulceration, bleeding, tissue destruction, functional impairment and cardiac overload [1-4].

Treatment of AVMs consists of endovascular embolization, surgical resection or a combined approach. Total surgical resection is challenging in most cases due to critical location and infiltration into multiple tissue planes. Therefore, endovascular embolization is an alternative treatment to reduce circulation of the nidus and prevent the development of collaterals. The mechanism of embolization is

to form a thrombus containing erythrocytes and to regress the lesion by constricting and re-canalizing the vascular structures [2]. However, local and systemic complications, including superficial ulcers, deep necrosis, cutaneous fistula, pigmentation, acute kidney disease, rhabdomyolysis, and pulmonary embolism may occur after embolization [5-7].

In this study, the management of an AVM case that developed large cutaneous necrosis after ethylene vinyl alcohol copolymer (EVOH, Onyx®, Irvine, CA, USA) embolization is presented.

Case presentation

A 32-year-old male patient presented with painful mass and necrotic ulcer on the left gluteal region. The painful mass appeared in childhood, progressed and expanded during adolescence, and was diagnosed as Schobinger stage-2 AVM six months ago. EVOH embolization was applied by an interventional radiologist twice in two months interval. Five days after the second session, pain, discoloration, discharge and ulcer developed, respectively.

The physical examination revealed edema, inflammation and 8x10 cm deep and dry necrosis with discharge and foul smell in the posterolateral of the left gluteal and adjacent trochanteric region (Figure 1a).

Table 1. Lower Extremity Functional Scale (LEFS).

Questions*	**Pre-op**	**Post-op 12th month**
Any of your usual work, housework or school activities.	3	4
Your usual hobbies, recreational or sporting activities.	2	4
Getting into or out of the bath.	3	4
Walking between rooms.	3	4
Putting on your shoes or socks.	3	4
Squatting.	1	3
Lifting an object, like a bag of groceries from the floor.	3	4
Performing light activities around your home.	3	4
Performing heavy activities around your home.	2	3
Getting into or out of a car.	3	4
Walking 2 blocks.	3	4
Walking a mile.	2	4
Going up or down 10 stairs (about 1 flight of stairs).	2	4
Standing for 1 hour.	2	4
Sitting for 1 hour.	2	3
Running on even ground.	3	4
Running on uneven ground.	2	4
Making sharp turns while running fast.	2	3
Hopping.	2	3
Rolling over in bed.	3	4

Score 0: Extreme difficulty or unable to perform activity, Score 1: Quite a bit of difficulty, Score 2: Moderate difficulty, Score 3: A little bit of difficulty, Score 4: No difficulty.

Figure 1a-c. Pre-operative (a), intra-operative (b), and post-operative 12th month (c) images.

There was no sign of systemic complication. The patient had difficulty in daily and work activities. The mean score of patient's Lower Extremity Functional Scale (LEFS) was 2.45 ± 0.6 (Table 1).

In laboratory tests, anemia, neutrophilic leukocytosis, increased infection parameters were observed (Table 2). Magnetic resonance imaging (MRI) examination revealed an enlarged tortuous vascular nidus, ischemic and infected tissues measuring 108x35 mm in the subcutaneous tissue of the left gluteal region and 55x42 mm in the left gluteal muscles (Figure 2a, b).

Table 2. Laboratory results.

Laboratory test	Initial results	Follow-up results
Hemoglobin (g/dl)	11.2	14.3
WBC (10^3/uL)	26.460	6.630
Neutrophil (%)	% 79.7	%55.1
Thrombocyte (10^3/uL)	359	403
CRP (mg/dl)	21,09	1,98
Procalcitonin (ng/mL)	0,61	0,01
Glucose (mg/dl)	103	95
Albumin (g/dl)	4.1	5.2
AST (U/L)	48	15
ALT (U/L)	36	11
Creatinine (mg/dl)	0.85	0.74

After obtaining patient's consent, all necrotic tissues were debrided, the vascular lesion was excised as much as possible, and the bleeding was coagulated and compressed with purse-string sutures (Figure 1b). During the debridement, we observed the musculocutaneous perforators of the necrotic skin derived from gluteal arteries to have occluded. The specimen was sent for histopathological examination. The defect was closed with advancement of wound edges

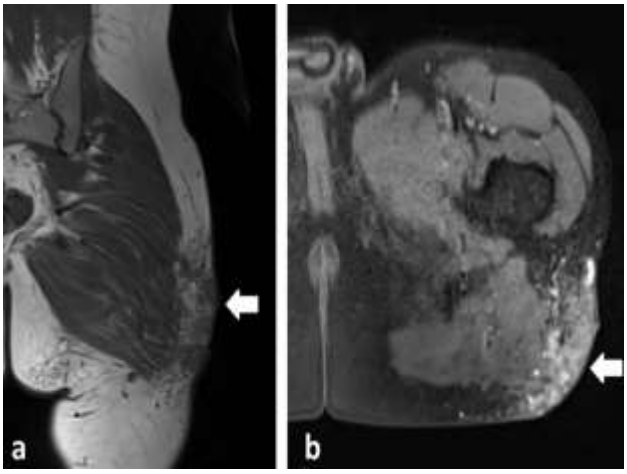

Figure 2. Pre-operative coronal (a) and axial (b) images of the MRI.

(Figure 1c). Penrose drain and compression garment were applied. The patient was discharged with oral analgesic and empirical antibiotic on post-op first day. Histopathological evaluation reported epidermal necrosis, diffuses hemorrhagic infarct, subcutaneous cavernous and thrombosed vascular structures and inflamed fibro-adipose tissues (Figure 3).

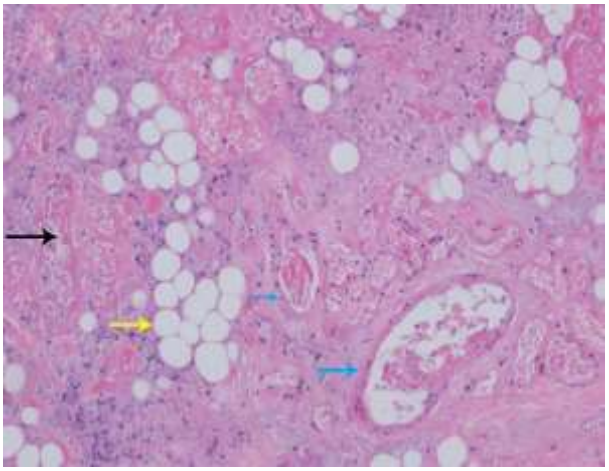

Figure 3. Histopathological imaging of the specimen. a) Necrosis and hemorrhagic infarct (black arrow), necrotic fat tissue (yellow arrow) and thrombosed vascular structures (blue arrows).

The patient was followed for approximately 19 months. The surgical area healed without

complications. The mean LEFS score was significantly improved to 3.75±0.44 score in 12 months postoperatively ($p < 0.00001$, the paired sample t-test). The patient had no significant functional limitations affecting work and daily life.

Discussion

Endovascular embolization has been the first treatment option in deep-seated forms of AVMs where the parenchymal component is low, the fistula between the artery and vein is prominent, and surgery is not possible [5]. Various embolizing agents including ethanol, bleomycin, n-butyl cyanoacrylate, and EVOH are used in the treatment [1,5,7]. EVOH is one of the most commonly used non-adhesive liquid embolizing agents with effective and safe treatment. EVOH provides more controlled devascularization with longer polymerization time, filling the AVM more completely, and lower tendency to migrate outside of the target site compared to other embolizing agents [8,9].

Complications after EVOH embolization are not rare, but the majorities are local complications. Schmidt et al. reported an 8.5% peri-operative complication including non-target embolization and ineffective treatment, and 31.5% post-operative complication including prolonged pain, persistent skin darkening, local infection, skin necrosis and finger amputation after 35 EVOH embolization in 15 peripheral AVMs [8]. Albuquerque et al. reported 16% complication including 3 skin necrosis and 1 active bleeding after 25 non-adhesive liquid embolizing agents with EVOH (Onyx®) and Squid® in 14 peripheral AVMs [10]. Saeed et al. reported 25% complications including embolic stroke, severe pain and bradycardia, catheter fracture, dark skin discoloration, arterial spasm, reflux to brachial artery after 28 EVOH embolization of 19 peripheral AVMs [9].

Managing embolization-related local complications of AVMs is easier than non-embolization complications. Although embolization procedures to the AVM create microthrombi within the vascular bundle, it also causes necrotic changes in the tissues fed from that vascular network [2]. Even if these necrotic tissues are debrided with surgical intervention, the dense vascular network consistent with the underlying vascular malformation shows a high bleeding tendency. In order to provide full control of bleeding during the operation, closure of the tissue with pledgeted or purse-string sutures provides significant benefit [4]. In our case, gluteal muscular perforators nourishing the skin angiosome around the AVM became occluded after embolization, and large skin necrosis developed. However, obliteration of this vascular network resulted in less bleeding than expected during debridement and excision of the AVM.

Not all embolization-related skin necrosis requires emergency surgical debridement because in some cases of skin necrosis that do not breach the deep dermis, de-epithelialization may occur, and the wound may heal spontaneously through secondary intention thanks to the widespread collateral vascular network in the AVM area. However, if the necrotic area becomes more defined and healthy tissue from below does not fill the defect, this should prompt consideration of the need for surgical debridement.

Conclusions

In conclusion, the post-embolization color changes in the skin should raise suspicion of skin necrosis, and after a diagnosis of skin necrosis is established, surgical treatments should be considered. Surgical treatment is indispensable both in the treatment of AVMs and in the management of embolization-related complications.

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