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Case Report

Torrential bleeding of arteriovenous malformation in hand post-ethanol sclerotherapy: A case report ☆

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ABSTRACT

Embolization or sclerotherapy is considered as the first-line therapy for the management of arteriovenous malformations (AVM) and can be performed directly targeting the nidus. Ethanol is an effective embolic agent; however, some complications may arise. This paper illustrates a case of torrential bleeding following ethanol sclerotherapy in a patient with progressive hand arteriovenous malformations with a poor prognosis and was suggested to be amputated. Direct pressure, tourniquet appliance, and split-thickness skin graft procedure were performed to stop the bleeding successfully. No recurrence of bleeding was reported; and complete alleviation of pain was achieved.

Introduction

Embolization is considered as the first-line therapy for arteriovenous malformations (AVMs) [1]. Other treatment options include pharmacological, surgical, and radio surgical approach. These treatments could produce a more beneficial outcome in certain circumstances, but embolization becomes the preferable treatment option as it provides acceptable nidus occlusion and a lower rate of both complications and recurrence. [2,3] Disease classification, which is based on angioarchitecture of AVM [4,5], provides embolization approach options that could help formulate the treatment for the patient.

Absolute ethanol is a common liquid embolic agent used to treat vascular malformation, despite its known toxicity because it is very effective in occluding vessels [6]. Dosage adjustment is highly recommended as major and minor complications could arise from its use [7]. Although rare, major complications such as limb amputation and cardiovascular collapse may take place [8]. Torrential bleeding was not reported as one of the major complications following ethanol sclerotherapy [6]. Minor complications may include local bullae, local tissue necrosis, and transient local neurological

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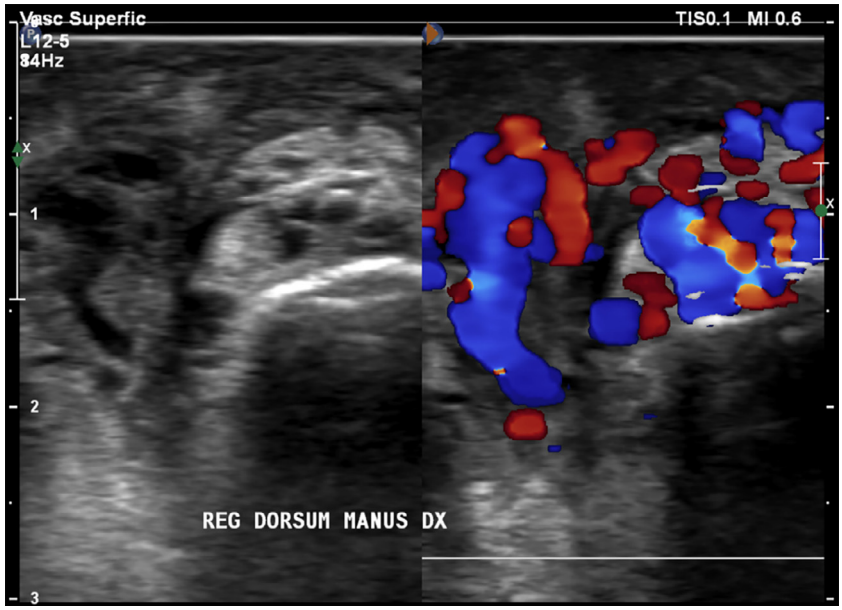

Fig. 1 – Initial Doppler ultrasound examination. Doppler ultrasound identifies the vascular nidus, with a "bag of worm" appearance, at the dorsal region of the right hand.

deficits [9–11]. This report illustrates a case of torrential bleeding following ethanol sclerotherapy in a patient with progressive hand AVM with a poor prognosis and was suggested to be amputated. Nevertheless, the patient provided consent to possible emergency amputation.

Case Report

A 27 years old male was introduced with a mass on the right palm involving the third and fourth digits. The lesion started to develop 20 years before his visit and had progressed rigorously for the past 1 year. The patient had a history of surgical mass removal 11 years ago and claimed that the mass was no longer visible after the surgery. The mass then slowly grew back in size accompanied by pain (numeric rating scale) reached 8 in the ring finger for 10 years afterward. Subsequently, the tip of the ring finger developed a necrotic wound, which was spreading slowly proximally. A surgery by another team was followed to debride the necrotic tissue. The first computer tomography angiography (CTA) examination showed AVM with multiple nidi at third and fourth digits with feeding arteries from the radial and ulnar artery and was confirmed under the Doppler ultrasound examination. (Fig. 1)

The patient then underwent the first digital subtraction angiography (DSA) of the right hand, followed by sclerotherapy using absolute ethanol and iohexol solution for injection (Omnipaque, GE Healthcare, Shanghai, China) with 8 to 2 ratios via direct puncture. Sclerotherapy targeted the nidi between third- and fourth-hand rays at the palm region. Posttreatment DSA showed some devascularization on the nidi (Fig. 2) Clinical symptom was alleviated with a significant reduction of pain (numeric rating scale 2). After the embolization, spontaneous bleeding occurred in the distal fourth finger. Direct pressure at bleeding site and finger tourniquet were applied to stop the bleeding. Surgical hemostasis and debridement procedure were performed on the fourth fingertip.

Second sclerotherapy guided by ultrasound and fluoroscopy using 20 mL of absolute ethanol and Omnipaque (GE Healthcare, Shanghai, China) mixture was performed in the following month. One day after sclerotherapy, multiple bullae appeared at the main sites of injection. (Fig. 3) Drainage of multiple bullae was performed on the patient's right hand. The patient reported complete alleviation of pain following the procedure.

Incidentally, spontaneous massive bleeding occurred at the same site as the location of vein varix during guided sclerotherapy 3 weeks after (Fig 4, supplementary video available on online version). The bleeding was torrential, continuous arterial bleeding, and unresolved with direct pressure using layers of gauze. It was managed by pressure dressing under the temporary application of tourniquet (only during dressing for 5 minutes). Constant direct pressure with the elastic bandage was applied successfully to prevent rebleeding. Following this occurrence of torrential bleeding, patient's hand was planned to be amputated, however the case was postponed until further angiography evaluation. Subsequent dressing changes were conducted once every week using the same technique of tourniquet application.

After one-month, multiple granulation tissues were formed. No bleeding occurred during a dressing change, even without the need to apply a tourniquet. DSA showed that the nidus was found to be much less in extension compared to previous DSA. (Fig. 5) Amputation procedure was decided as unnecessary and further limb-preserving management

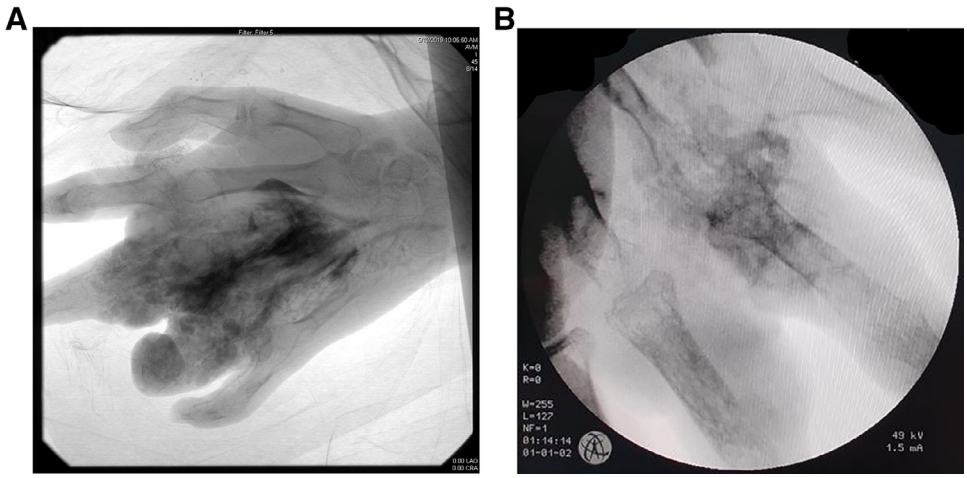

Fig. 2 – Angiogram after first and second sclerotherapy. A: First direct puncture sclerotherapy under DSA using absolute ethanol was targeted on the nidus. Post sclerotherapy angiogram shows adequate devascularization. B: Second sclerotherapy a month after the first sclerotherapy, guided by ultrasound and fluoroscopy, targeted the nidus around third and fourth digits until devascularization was visualized.

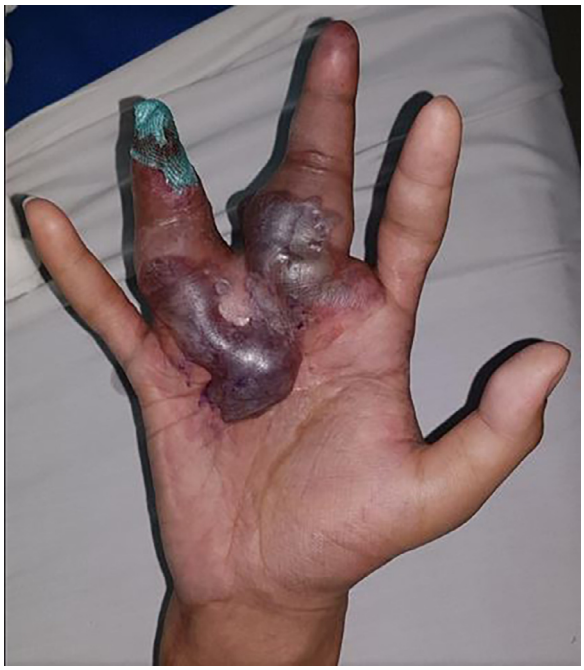

Fig. 3 – Multiple bullae after the second sclerotherapy. One day after the second sclerotherapy, multiple bullae developed as seen on the volar side.

was conducted. Split-thickness skin graft (STSG) was performed to prevent further bleeding complications and cover the granulation tissue. The graft worked well. Fig. 6 shows 3 weeks after surgery. The patient was scheduled for additional sclerotherapy while embarking on passive and active range of motion exercise for his thumb, index, and little finger. All procedures were performed with the patient's consent.

Discussion

AVM may occur in any body part, which barely easy to identify at its early stage [11]. It is evident in the mass of the right palm which was detectable when the patient was already 8 years of age. Typically, AVM is staged with Schobinger classification [11,12]. In this case, an upstaging from stage II to stage III was depicted when the patient started to suffer from severe pain, and the fourth fingertip developed a necrotic wound. This progressivity and necrosis are associated with arterial steal syndrome of the AVM that reduces tissue nutritive flow [13].

AVM is diagnosed primarily by ultrasound and magnetic resonance imaging (MRI). Ultrasound is useful for analyzing flow patterns to diagnose fast-flow malformations and to confirm arteriovenous shunting shown in the patient [12,14]. Dynamic MRI is useful for identifying inflow arteries, outflow veins, and the location of shunting. Despite its limited utilization compared to dynamic MRI, CTA with high spatial resolution could be beneficial, as in this case, to map feeding and draining compartments and evaluate bony involvement [14]. CTA of the patient showed complex AVM, with extensive involvement of the palm and the third and fourth digits, with cortical erosion of the mid-phalanx of the fourth digit. Multiple dilated shunts, identified as a complex vascular network, and dilated veins in CTA were confirmed by conventional arteriography before the first sclerotherapy.

The AVM should be further classified according to its angiographic pattern, as could be seen by CTA. The presence of multiple dilated shunts corresponds to type IIIb of Cho-Do classification and type IIIb of Yakes classification lesion [12]. This angiographic pattern classification would be needed to determine further treatment. Then, combined with the Schob-

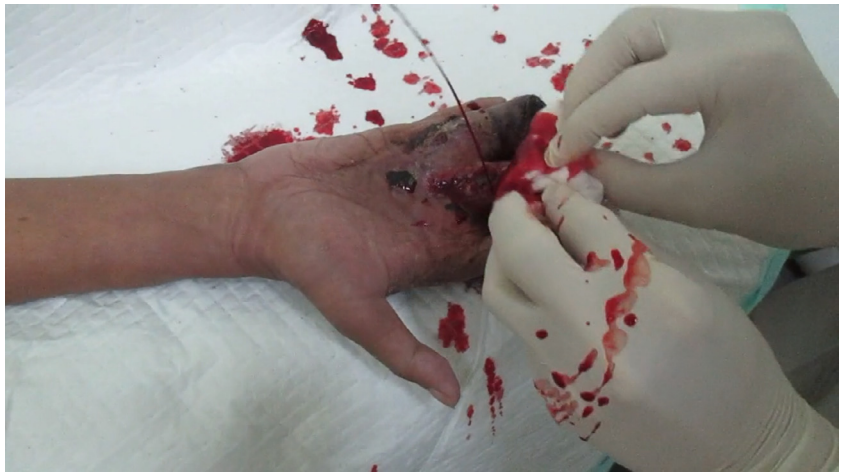

Fig. 4 – Torrential bleeding during guided sclerotherapy three weeks after second sclerotherapy. The bleeding was torrential, continuous arterial bleeding, and unresolved with direct pressure using layers of gauze. It was managed by pressure dressing under the temporary application of tourniquet (only during dressing for 5 minutes). Constant direct pressure with the elastic bandage was applied successfully to prevent rebleeding. [JP1] [JP1] Added additional picture to represent the torrential bleeding occurred. (Including a still image from the video (at about 5 sec into the video) would be helpful for those who do not want to download and view the entire video).)

inger classification, as in the presented case, percutaneous sclerotherapy, endovascular embolization, or surgery would be indicated [12,13,15]. However, the surgical approach alone may cause life-threatening bleeding and also could result in AVM explosive growth due to incomplete resection [15]. Therefore, curative embolization or as preoperative devascularization before surgery [15,16] was the concern for this case.

Among the 3 liquid agents commonly used, ethylene-vinyl-alcohol-copolymer and n-butyl cyanoacrylate has less local and systemic complication than ethanol [17]. Ethanol was chosen because it was more cost-effective than the others in regards to this case and yielded a high response rate, yet the frequent complications [6]. Direct puncture injection of ethanol is also referred to as ethanol sclerotherapy due to its sclerosing properties [14,18]. Ethanol sclerotherapy was most effective in type II and IIIb of Cho-Do classification, with complete occlusion rate of 83% [4].

Hemorrhage in AVM could develop due to its natural progression or as a posttreatment complication. During its natural course, as the AVM enlarges, the inability to adjust the blood flow would weaken the vascular wall. These areas are weak points with a high rate of bleeding and should be eliminated by endovascular treatment [19]. Nevertheless, if hemorrhage occurs after endovascular treatment, bleeding due to posttreatment complications should be considered. Ethanol toxicity could cause transmural vascular necrosis extending to superficial skin and increases the risk of hemorrhage [18]. In high flow malformations, dilution of the ethanol could also result in unintended infarctions of tissue downstream [20]. Should venous outflow is compromised or thrombosed partially during embolization while the arterial blood supply is preserved, the increase of shear stress would also increase the risk of hemorrhage [20]. Such examples could be seen in the case with high flow bleeding 1 month after the second scle-

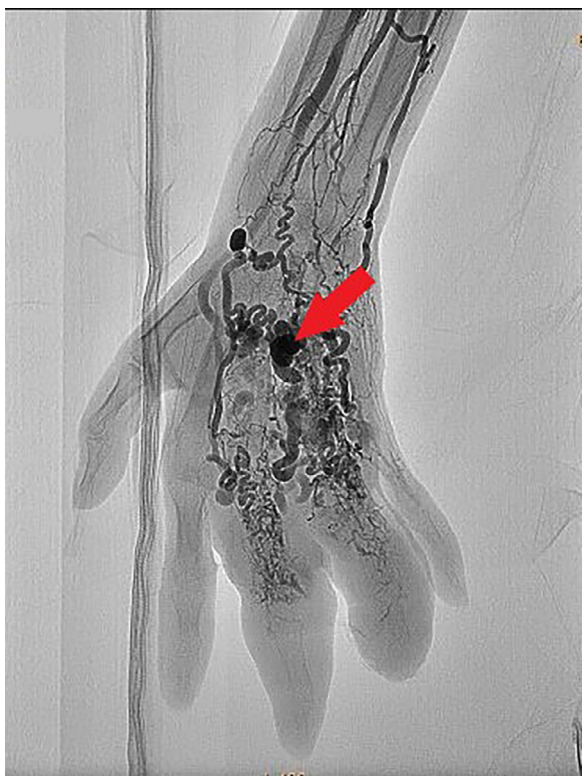

Fig. 5 – Follow up evaluation by DSA one month after second sclerotherapy. Multiple tortuous vessels, indicating vascular nidus, were visualized around the metacarpal region, multiple feeding arteries, from the radial artery, interosseous artery, and ulnar artery was also identified. Vascular nidus was found to be smaller in size (red arrow).

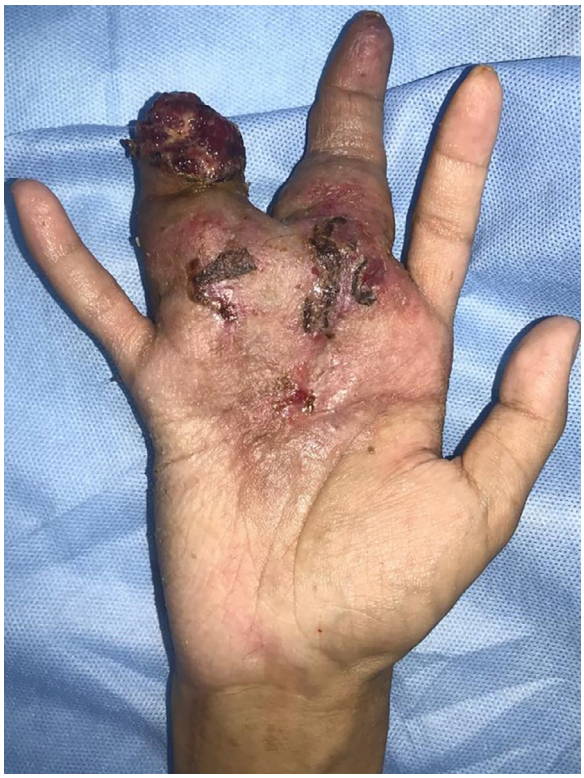

Fig. 6 – The wound three weeks after the STSG procedure. The graft takes, no torrential bleeding occurred. A minor raw surface on the tip of the forth digit heals secondarily.

rotherapy. As the location of bleeding is in the same site of venous varix identified previously, which was punctured during treatment, hemorrhage of this patient most likely related to endovascular treatment, mainly due to ethanol transmural thrombosis.

Hemorrhage is an absolute indication of prompt treatment for AVM [21]. Nevertheless, in our case, emergency endovascular treatment could not be performed. Without a doubt, treatment by tourniquet use along with direct pressure, and constant pressure with the elastic bandage was effective to curb the emergency problem. Despite the plan for further treatment, the hemorrhage was found to stop completely, with AVM nidi found to be smaller in size compared to previous DSA. While tourniquet and direct compression by itself are a bleeding control method, the complete cessation of the bleeding would also be due to the effect of the delayed sclerosing activity of ethanol 1 month before the vascular damage process [14]. A transient posttreatment inflammatory response has been reported in this case, in which full response to the treatment could be evaluated.

In the final presentation of the case, the hemorrhage has stopped with no more complaint of pain being reported. Wound dressing followed by split-thickness skin graft was undertaken to cover granulation tissue and prevent further complications. In consideration of the case, multimodality treatment was administered as it is crucial for high-flow hemorrhage following AVM sclerotherapy. Apart from the necessity to treat further the AVM pathology, the technique reported here had been successful in managing the high flow of hemorrhage. While complete pain reduction was achieved, the patient was not completely healed from the AVM and was scheduled for further treatment sessions.

This case showed the potential of high flow hemorrhage following AVM ethanol sclerotherapy which then later showed complete cessation with constant pressure using elastic bandages. This case also showed ethanol sclerotherapy clinical benefit in alleviating pain completely. It would encourage ethanol sclerotherapy for AVM despite the fear of its side effect, which could be treated by proper pressure appliance using temporary aid of tourniquet.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2020.06.017.

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