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CASE REPORT

# Case Report of Ethanol and Cyanoacrylate Embolisation of a Recurrent Uncontrollable Torrentially Bleeding Arteriovenous Malformation of the Finger

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**Introduction:** In general, arteriovenous malformations (AVMs) are extremely rare, with an incidence of only 1 in 100,000. They are rarer still in the hands and present variably with bleeding, heaviness, a pulsatile mass, pain, ulceration, or necrosis.

**Report:** The case of a 25 year old man with a rapidly bleeding right thumb AVM is presented. Bleeding was torrential and life threatening within a matter of seconds. He had previously undergone surgical ligation and embolisation twice at another centre, without success. At presentation, he had no thumb function and the bones of the thumb were exposed. An angio-embolisation was performed with ethanol and cyanoacrylate as the embolic agent. This was done using direct puncture into the AVM and also with a transarterial approach with microcatheters inserted into various unnamed branches feeding the AVM. Non-target embolisation and reflux was prevented by deploying a pneumatic tourniquet and mechanical elastic bands to confine the flow of the embolic agents within the AVM. Re-aspiration of the embolic agent post-embolisation was also performed to prevent local/systemic ethanol toxicity. Haemostasis was achieved without the need for further compression. A right thumb disarticulation was subsequently performed and the patient expressed great satisfaction with the outcome.

**Discussion:** AVMs in the hand are particularly challenging to treat owing to the need to preserve function of the myriad tissues and structural units that enable the many hand movements involved in activities of daily living. Even a partial loss of function may be disabling or poorly tolerated. The mainstays of treatment are embolisation, sclerotherapy, and surgical ligation/resection, all of which carry the potential for ischaemic injury to muscle and soft tissue. A holistic approach to management is desirable prior to selecting the appropriate management plan.

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## INTRODUCTION

Arteriovenous malformations (AVMs) are usually present at birth; however, not all of them are easily detectable clinically and can be affected by trauma, surgery, or hormonal alterations. They can occur on any part of the body, including the hand, which is the second most commonly affected site.[1]

The treatment of hand AVMs includes conservative treatment, embolisation/sclerotherapy, surgical ligation, excision, and amputation. However, there is currently no consensus on treatment.

A case of a young man who was diagnosed with a congenital thumb AVM who failed initial treatment is reported. He subsequently underwent a repeat angio-embolisation, which successfully stopped the torrential AVM bleed.

## CASE REPORT

A 25 year old man suffered swelling and bleeding from his right thumb. He lost the use of his thumb as he constantly required a heavy compression dressing to arrest the torrential haemorrhage (Fig. 1a). Despite previous ligation of his radial artery and multiple rounds of embolisation therapy, the bleeding was still not controlled.

At presentation, there was torrential, life threatening arterial bleeding, necrosis with exposed bone, and macerated skin from prolonged bandaging (Fig. 1b), as well as symptomatic anaemia. Given his late stage presentation, hand surgeons were insistent on a hand amputation.

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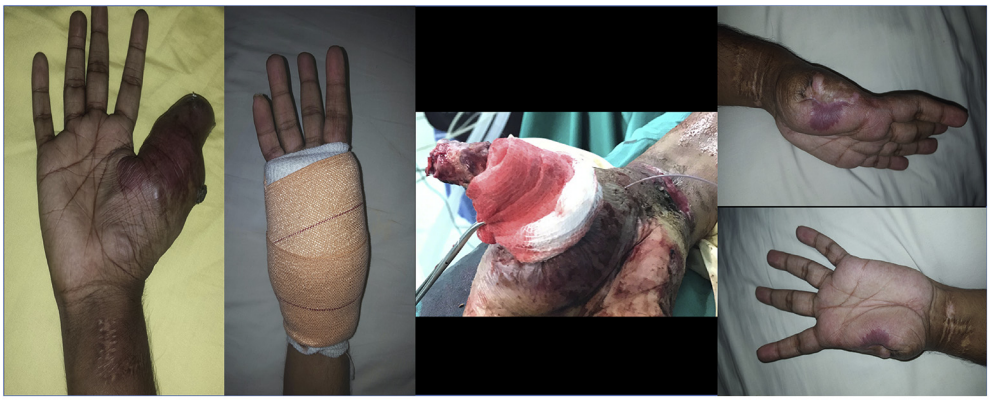

a b c

**Figure 1. a.** First presentation of AVM in 2014. The patient had open ligation of his radial artery and as well as embolization performed. Despite surgical and embolization therapy, bleeding was poorly controlled, requiring prolonged bandage over the right thumb as seen in the above picture. **b.** Subsequent presentation of AVM in 2017. Severe skin necrosis with bone exposure and torrential bleed were noted. **c.** 6 months post surgery with good wound healing and cessation of bleeding.

However, in view of the clinical urgency and the patient's social circumstances, further angio-embolisation was selected with the aim of securing definitive haemostasis, digit preservation, and the hope of preventing further recurrence.

The procedure was performed under general anaesthesia with a flat panel system (Artis Zeego, Siemens AG, Forchheim, Germany). The right brachial artery was cannulated in an antegrade manner under ultrasound guidance and a 4 Fr sheath was inserted. Baseline angiography was obtained for evaluation of the AVM and for selection of access route, transarterial, direct puncture, or both. A transarterial approach via the ulnar artery was performed using a 0.018 inch Terumo wire (Terumo, Somerset, NJ, USA) and a BerII catheter (Cordis, Miami Lakes, FL, USA). Diagnostic angiography revealed a previously ligated radial artery, feeding branches from the ulnar artery and palmar arches (Fig. 2a), as well as large draining veins (Fig. 2b).

Using the direct puncture method, a 21 G needle was directed into the prominent draining veins, checking backflow of blood by gentle aspiration (Fig. 3a). To determine the volume and injection rates of ethanol, test injections of contrast media were used to fill the AVM without opacifying normal vessels. With a tourniquet (A.T.S 2000; Zimmer, Warsaw, IN, USA) inflated at 250 mmHg and mechanical elastic constrictors around the proximal phalanges to achieve vascular stasis (Fig. 2c), ethanol was injected with Lipiodol (Guerbet, Villepinte, France) into the AVM (Fig. 3b). Five minutes after ethanol injection, an angiogram was performed to evaluate the response to embolisation. However, a post-embolisation angiogram revealed suboptimal results with persistent contrast filling of the AVM (Fig. 3c).

The decision was made to repeat the embolisation with 20% cyanoacrylate glue (B. Braun, Melsungen, Germany) by mixing 2 mL lipidiol and 0.5 mL glue together with ethanol (Fig. 3d). A 2.7 Fr microcatheter (Terumo) was then superselectively advanced via the digital artery of the index finger to be within close proximity of the new nidus. Multiple runs of glue and ethanol were performed to achieve best results, with careful manipulation and repeat aspiration of remnant ethanol post-embolisation (Fig. 3e). A significant reduction in bleeding was achieved following these manoeuvres. In total, 10 mL ethanol and 0.5 mL cyanoacrylate glue was used.

A completion angiogram revealed no further collaterals to the AVM (Fig. 3f). The brachial artery sheath was removed and haemostasis was achieved by manual compression of the brachial artery puncture site.

Thereafter, the patient underwent a right thumb disarticulation 3 days later. He had recovered well 8 months post-surgery (Fig. 1c). He was able to regain good hand function, for example in driving and performing activities of daily living. His quality of life has significantly improved and the team is most happy with the outcome. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

## DISCUSSION

AVMs are congenital vascular malformations that can occur in any part of the body and may not be apparent in the early stages, especially on the digits. They are typically staged based on the Schobinger staging system and classified accordingly to the classification by the International Society for The Study of Vascular Anomalies.[2,3]

Management of hand AVMs has been exceptionally challenging because of the functional complexity strictly related to the anatomy where many different tissues of high functional value are located in a small space. In digital AVMs, treatment is often indicated, especially so when the AVM involves the thumb, arguably the most important digit for finger opposition and hand function.

Endovascular techniques have been used in an attempt to maintain function without permanent disability or recurrence from reconstituted arterial flow from the nidus.