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Case report

Diagnosis and management of calcaneal Yakes type IV AVM: Two case reports ☆

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ABSTRACT

Peripheral arteriovenous malformations (AVMs) are rare, congenital anomalies resulting from developmental genetic defects of the vasculature and cause significant disturbances in daily activities. AVMs can affect any part of the body and are rarely observed in the calcaneal region. Here, we describe two cases of Yakes type IV calcaneal AVMs managed by ethanol sclerotherapy at our institute.

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Peripheral arteriovenous malformation (AVM) is a rare disease, especially in the calcaneal region. Although previous publications have reported AVMs in the foot, AVMs in the heel are uncommon, and few have been described as Yakes type IV AVMs. Peripheral AVMs are diagnosed according to clinical symptoms and medical imaging, such as doppler ultrasound, multislice computed tomography, magnetic resonance imaging (MRI), and angiography, which is the current gold standard. Cho et al. established a classification system for AVM in 2006, and Yakes proposed modifications in 2017, both of which utilized angiography to interpret the angioarchitecture of malformations for endovascular management and follow-up [1,2]. Cho's classification system included 4 types: type I, fewer than 3 arteries shunting into a single drainage vein; type II, multiple arterioles shunting onto a single venous component; type IIIa, fine arteriolovenular shunting, observed as a blush or fine striation on angiography; and type IIIb: multiple arteriolovenular shunting that appears as a complex vascular network [2]. Yakes modified the classification to increase the comprehensibility and applicability of the system: type I, direct shunting between arteriovenous (AV) fistulas; type II, true "nidus," in which the components include direct arterio-arterial inflow to

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a venulo-venous structure; type IIIa, microfistulas from multiple arterio-arterial structures into a single enlarged, drainage vein; type IIIb, similar to type IIIa but involves multiple outflow veins; and type IV, innumerable microfistulous arteriolar structures shunting into innumerable venular connections that diffusely infiltrate a tissue [1].

The management of peripheral AVMs requires a multidisciplinary approach that can include endovascular, surgical, medical, and laser treatment. Endovascular management is currently considered the first-line and most effective treatment option. Absolute alcohol (99.5%) is highly effective for the treatment of peripheral AVMs, administered by direct puncture or trans-vessel injection. Absolute alcohol denatures the proteins on endothelial cells, inducing thrombosis in the injected vessel. Ethanol management increases the odds of curing peripheral AVMs but can also result in severe complications if ethanol reflux reaches normal vessels. Therefore, physicians must be appropriately trained in the administration of absolute alcohol for AVM management [3,4]. We report two cases of calcaneal Yakes type IV AVMs that were diagnosed and managed using ethanol at our hospital.

Case reports

Case 1

A 62-year-old woman was admitted to the hospital due to bleeding from an ulcer on the left heel. The patient had long noticed the presence of nodules, similar to warts, on her left heel that were painless and only experienced limited bleeding. The patient had previously undergone a minor surgical procedure for this presentation at a primary care hospital. However, the lesion became infected and necrotic and was being managed with antibiotics. The patient was referred to the Dermatology Department at the hospital and received laser treatment. The lesion became more severe, with increased ulceration, necrosis, and bleeding following the last treatment. On physical examination, an ulcerous, necrotic, pulsative lesion approximately 5 cm in size was identified on the left heel. Bleeding occurred when the scab fell off, which was stopped by compression (Fig. 1A). MRI of the left heel was hyperintense on T2-weighted images and short tau inversion recovery images with flow void, and a hyperintense signal with vascular structures was also observed on T1-weighted image fat-saturated (T1W FS) post-gadolinium (Fig. 1B and C). Angiography revealed a type IV AVM (Yakes classification) on the left heel, which was supplied by the posterior tibial artery and drained to the posterior tibial vein and plantar venous arch (Fig. 2). The patient was treated by endovascular embolization under general anesthesia through direct puncture with 3 24-G butterfly needles (B. Braun) at the nidus (Fig. 3). We injected a total volume of 10 mL 99.5% ethanol. Angiography after the procedure showed no persistent AVM nidus, and the healthy arterial and venous systems remained intact (Fig. 4). The lesion became purple and painful, with remaining ulceration. The wound was cared for using analgesic drugs and rinsed with warm saline daily. The patient experienced successful recovery at the heel and was able to walk normally 6 months after treatment (Fig. 5), with a satisfaction reported at 90% compared to before treatment.

Case 2

A 44-year-old woman with a history of heavy bleeding from the left foot was admitted to the hospital. The patient noticed a red macule on the left heel, which was painful when walking. The macule caused heavy bleeding 20 years prior and was treated with surgical hemostasis and sclerotherapy. The lesion became more severe during both of her pregnancies. On examination, an ulcerative lesion with uncontrolled bleeding was observed, despite compression. The patient underwent surgery to stop the bleeding and was transferred to the digital subtraction angiography (DSA) room for vascular investigation. The DSA images revealed an AVM in the left heel, which was supplied by the posterior tibial artery and drained to the posterior tibial vein and saphenous vein (Fig. 6). Three 24-G needles were used to puncture the nidus, and a total volume of 26 ml 99.5% ethanol was injected, together with compression of the drainage veins (Figs. 7 and 8).

Re-examination one month later revealed that the ulcer was healing, and the patient was scheduled for clinical assessment and MRI (Fig. 9). We performed percutaneous ethanol sclerotherapy directly to the nidus, using a total volume of 7 ml 99.5% ethanol. Post-treatment imaging indicated a significant reduction in nidus size (Fig. 10). Six months after treatment, the ulcer was perfectly healed (Fig. 10). The patient was relieved from resting pain, with only minor walking pain, and her satisfaction score was 9 of 10.

Discussion

Vascular malformations generally occur in a small percentage of the population, with an estimated incidence of 1.2% based on existing reports of neonatal congenital defects, and AVM represents the second-most common vascular malformation. AVMs most commonly occur in the central nervous system, but they can develop anywhere in the body, although they are rarely found in the foot region. The diagnosis and treatment of AVM remain challenging due to a high recurrence rate. Even in cases of radical treatment, the complication rate remains high, posing huge challenges for interventionists.

To develop a specific treatment plan, AVM is often classified into different types. In the past, the Schobinger classification system was the most commonly used, which was based on clinical evaluation, disease progression, and severity [5–8]. However, this classification system did consider imaging results, which are important considerations in describing the progression and severity of such anomalies. Therefore, the Schobinger classification system was preferred primarily by vascular surgeons for the assessment of treatment response.

With the rapid development of diagnostic imaging, angiography has become the gold standard for AVM diagnosis. Angiography can clearly visualize the feeding arteries, nidus, and drainage veins, contributing to the development of an optimal treatment strategy. According to this approach, Cho et al.

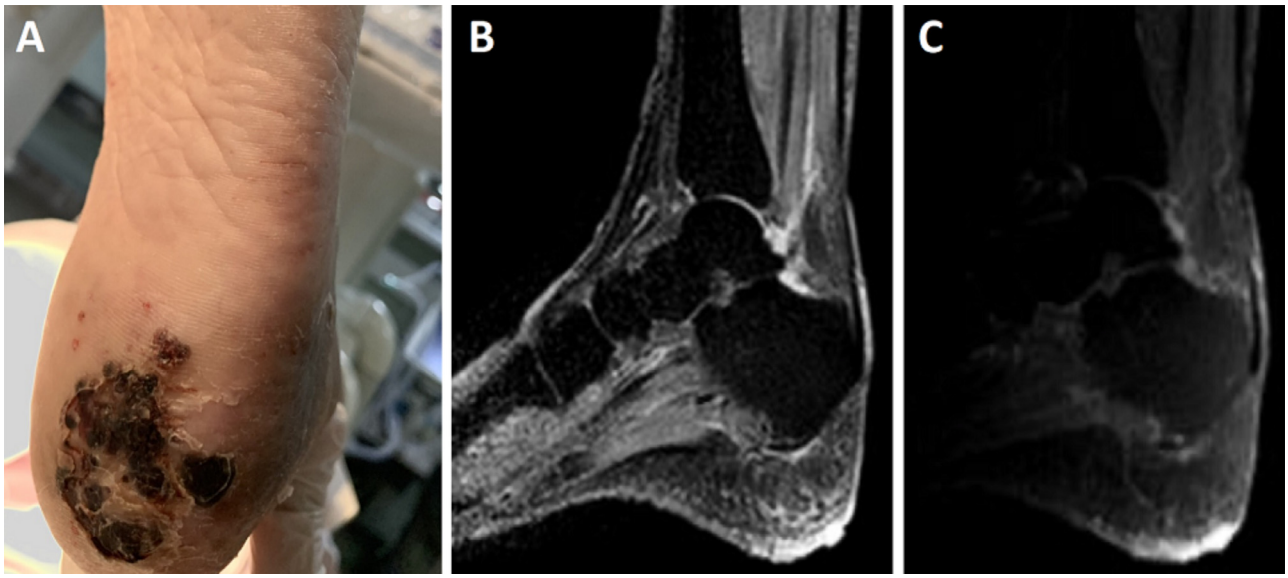

Fig. 1 – (A) A pulsative, necrotic, and painful lesion on the left heel. (B) Sagittal short tau inversion recovery (STIR) image was hyperintense with flow void structures. (C) A highly enhanced T1-weighted fat-saturated (T1W FS) image with gadolinium contrast showed the lesion with marked enhancement.

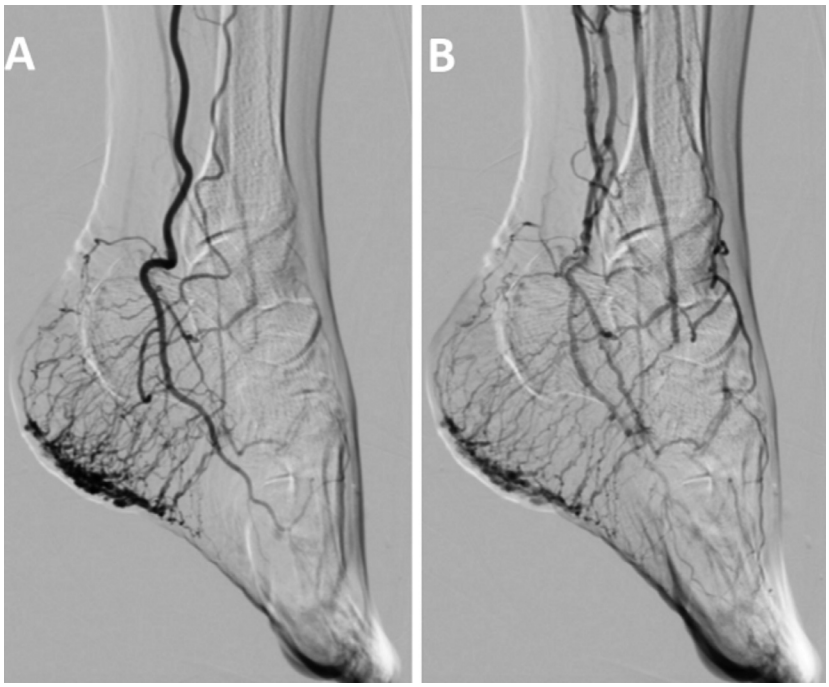

Fig. 2 – Angiography of the left heel showed a Yakes type IV AVM, which was supplied by branches from the posterior tibial artery (A) and drained to the posterior tibial vein (B).

(2006) and Yakes et al. (2017) defined a new AVM classification system.

Both classification systems are based on the nidus morphologies and drainage vein identification. The drainage veins are considered to be the primary cause of AVM development; therefore, treatment should focus on a venous approach or be targeted to a nidus located near the venous side. However, the Cho classification system does not include one malformation type that is commonly observed in the central nervous system. The Yakes classification system can be used to describe all AVM types, although Yakes defined direct AV fistulas as type I AVMs, which are considered simple malformation entities by the International Society for the Study of Vascular Anomalies 2014 guidelines [5,8].

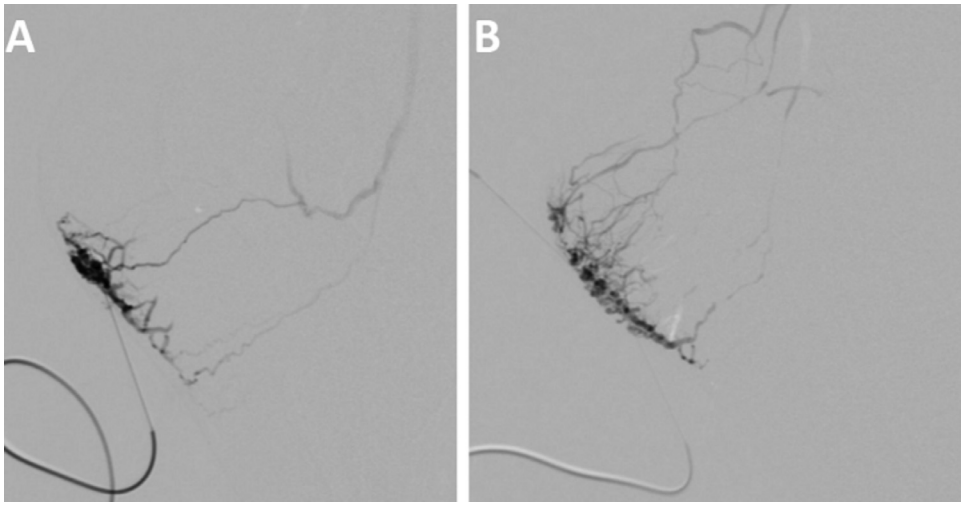

Fig. 3 – A high concentration of ethanol was injected through a direct puncture needle.

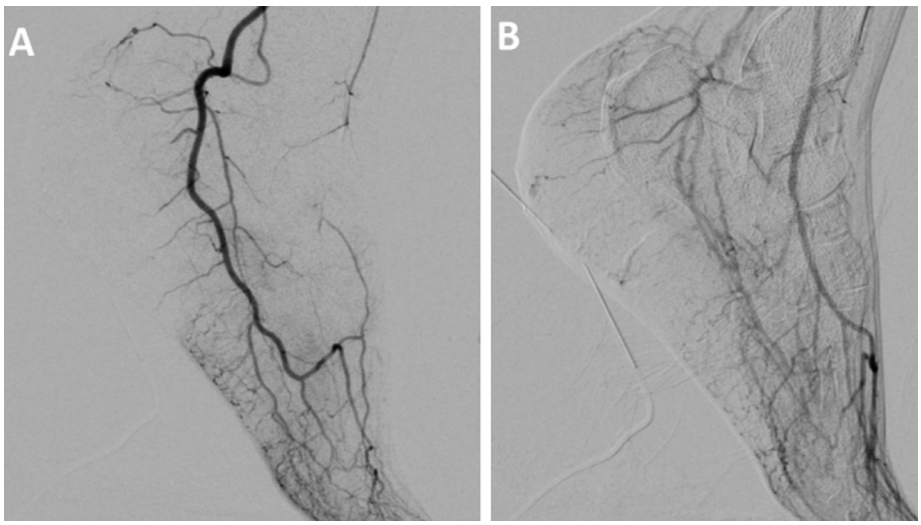

Fig. 4 – Re-examination angiography showed that the AVM was completely removed while retaining intact arteries and venous systems.

We applied the Yakes classification system to our treatment plan to precisely evaluate the lesion before the intervention. At our institute, we have treated multiple cases of peripheral AVMs, in addition to central nervous system AVMs; therefore, the Yakes classification is our preferred system for conducting diagnosis, treatment planning, and follow-up.

Our two cases of AVM are considered relatively rare. A retrospective report by Huyn described 29 cases of foot AVM over a 13-year period (1999–2012). However, the author did not describe the exact locations of those lesions.

Additionally, several case reports have described the occurrence of foot AVMs, which are commonly treated with alcohol sclerotherapy. The majority of foot AVM cases were described in the plantar aspect and classified as type IIIa or IIIb (Yakes classification) [9–14]. One case report described a Yakes type IV calcaneal foot AVM [4] featuring an ulcerated wound with a bleeding and pulsatile lesion, which were similar to the features observed in our cases.

Most reported cases have been treated with ethanol sclerotherapy, either through direct puncture or transarterial injection. In type III cases, a combination of ethanol and coiling is required to reduce the size of the drainage vein and reduce the required volume of ethanol, in accordance with the maximum allowable volume, which is 1 cc/kg. Moreover, in type IV AVMs, the accurate needle puncture of a microshunt location is necessary to allow the injected ethanol to eliminate the nidus, which typically requires not only a proper needle puncture technique but also drainage vein compression [2–4].

In our cases, drainage vein compression eliminated numerous microshunts. Although a small nidus remained on imaging, the patients' clinical symptoms improved without local complications. The ulcers were healed by soaking the feet in

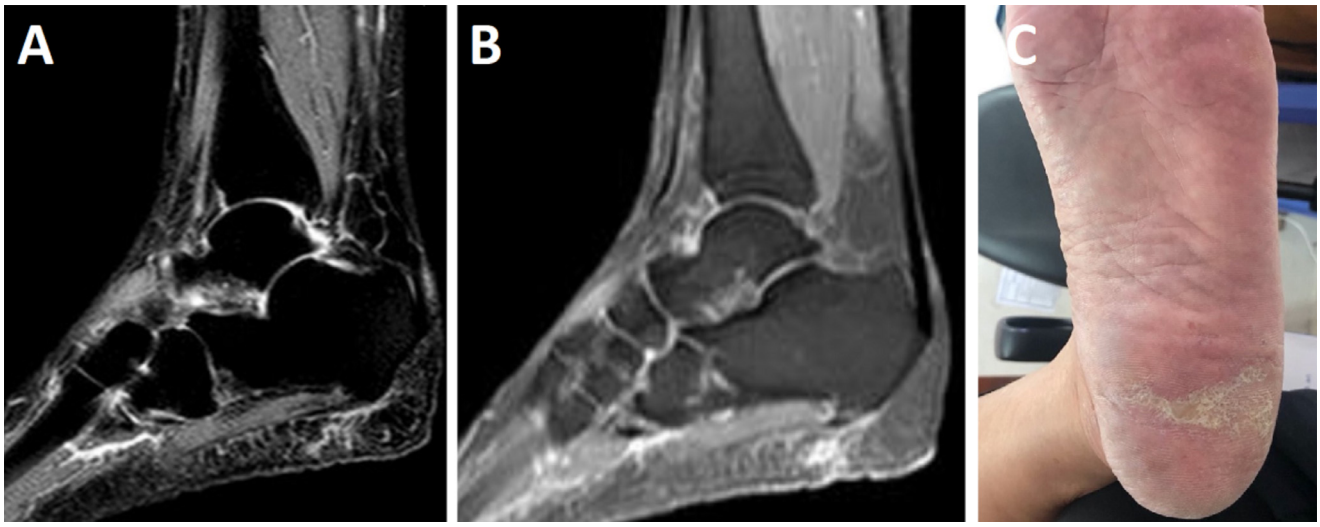

Fig. 5 – Follow-up MRI showed that the AVM was almost completely resolved (A and B). Heel skin post-treatment was normal (C).

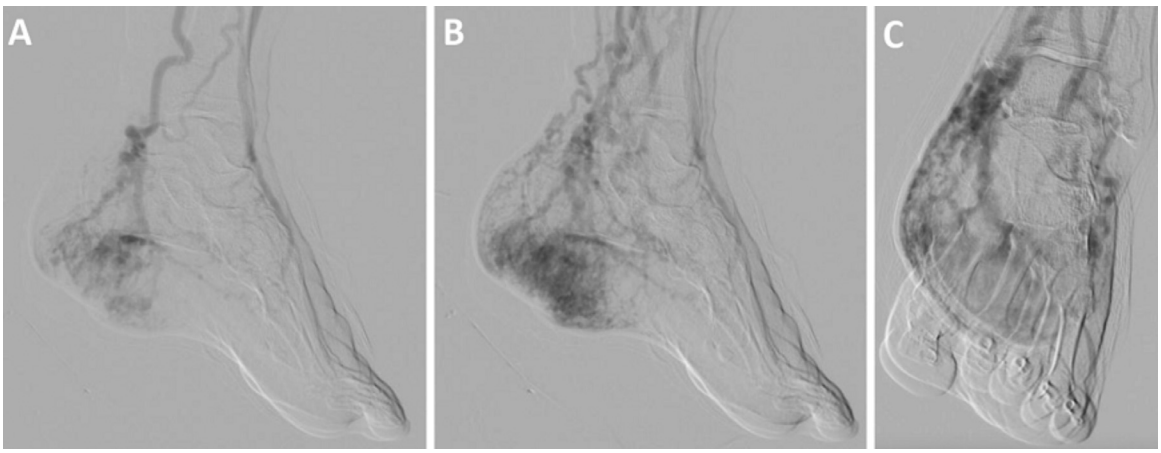

Fig. 6 – Angiography of the left foot revealed a heel AVM, fed by medial calcaneal branches and posterior calcaneal branches of the posterior tibial artery (A and B). Numerous microshunts were observed draining into the posterior tibial vein and saphenous vein (C).

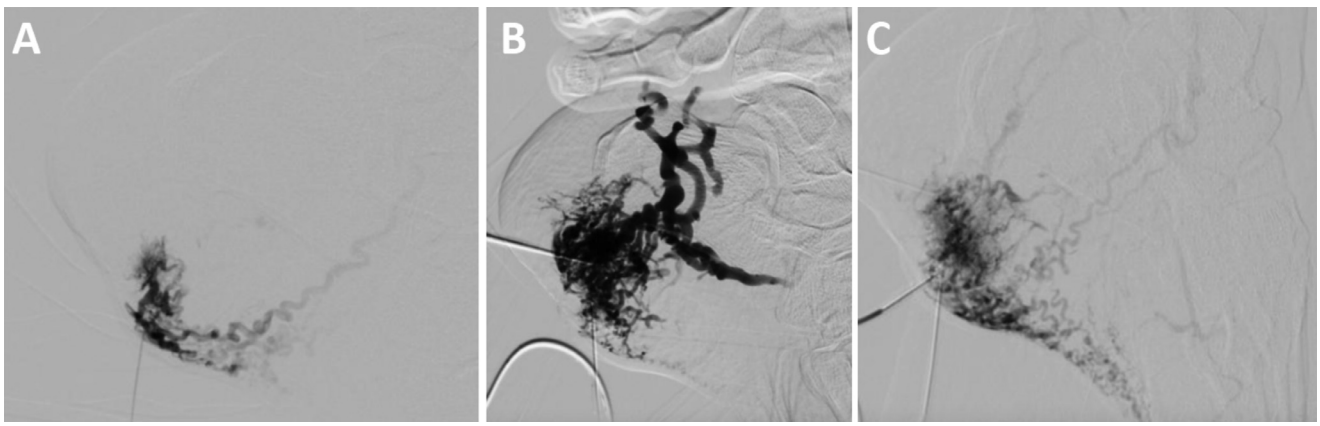

Fig. 7 – (A) Direct percutaneous angiography images showed microshunts between venules and arterioles. (B and C) Direct percutaneous puncture embolization with compression of the draining veins.

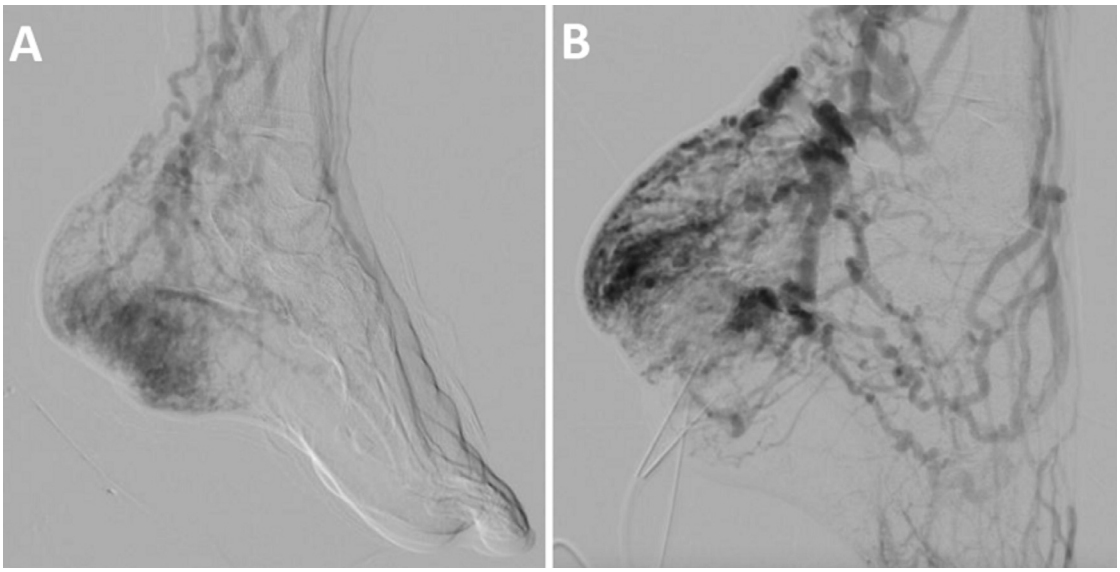

Fig. 8 – DSA images before and after treatment showed a clear reduction in the nidus size (A). A small remaining compartment was observed at the posterior heel (B).

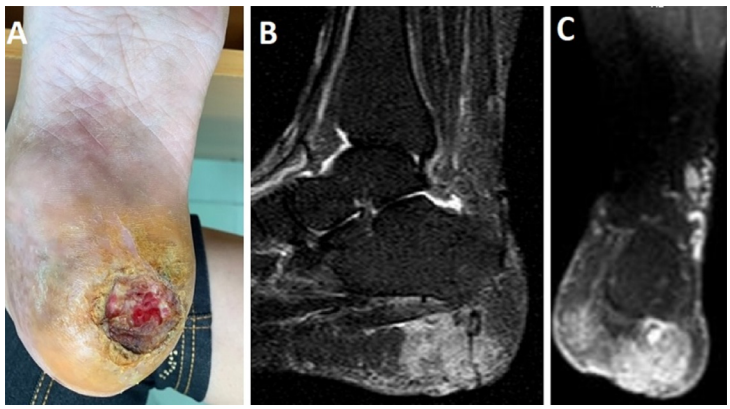

Fig. 9 – The ulcerative lesion in the left heel (A). The AVM nidus was hyperintense on short tau inversion recovery (STIR) imaging with a flow void (B), and homogeneous enhancement was observed on T1-weighted fat-saturated (T1W FS) post-contrast imaging (C).

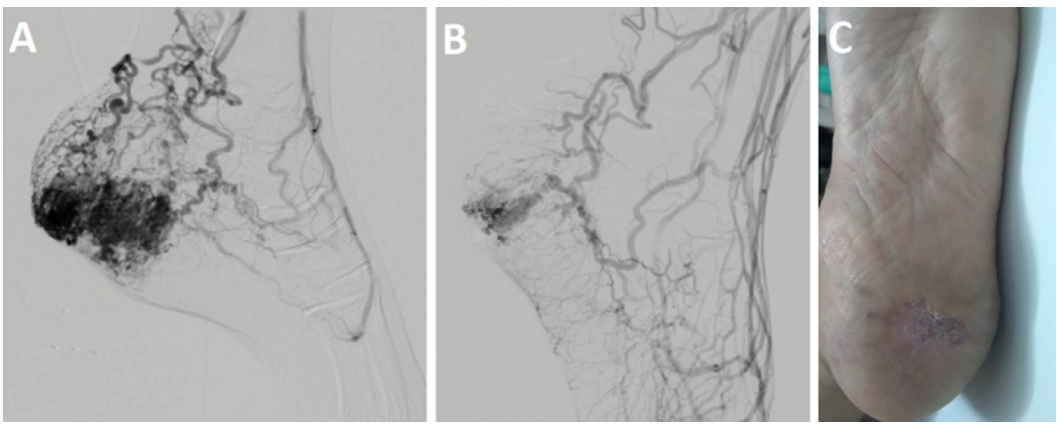

Fig. 10 – Angiography pre- and post-treatment revealed that the nidus was almost completely resolved (A). Six months after the second ethanol sclerotherapy treatment, the wound appeared completely healed.

warm water. Despite describing mild pain, both patients regained the ability to walk normally.

Conclusion

AVMs of the foot are rare clinical entities, especially Yakes type IV AVMs. Our two cases of type IV foot AVM were treated with ethanol injection, resulting in good clinical results with minor complications. These case experiences indicated that endovascular intervention using ethanol injection to treat type IV foot AVMs could result in positive outcomes. This embolotherapy technique can be combined with other embolization particles and drainage vein compression techniques.

Patient consent

Informed consent for patient information to be published in this article was obtained.

Ethical statement

Appropriate written informed consent was obtained for the publication of this case report and accompanying images.

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Author contributions

Nguyen DL, Tran H, and Nguyen MD contributed equally to this article as co-first authors. All authors have read the manuscript and agree to the contents.

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