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Case Report

Successful Complete Surgical Resection of a Large Venous Malformation of the Lower Extremity: A Case Report

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Venous malformations (VMs) are the most common type of vascular malformations, resulting from errors in vascular morphogenesis. Because of the wide variety in their presentations, selecting the appropriate treatment, especially for large VMs, may be challenging. Herein, we report a case of a 59-year-old man with a large VM in the lower extremity who achieved favorable outcomes by complete surgical resection. Even large VMs can be successfully treated with surgery when patients are properly selected. An accurate and careful evaluation is essential for achieving optimal outcome in patients with VMs.

Keywords: venous malformation, surgical resection, lower extremity

Introduction

Venous malformations (VMs) are the most common type of vascular malformations, resulting from errors in vascular morphogenesis. They can occur in any body part, but in approximately 40% of patients, they occur in the extremities. The clinical presentations of VMs vary based on their size and location and may include swelling or pain. Because of the wide variety in their presentations, selecting the appropriate treatment, especially for large VMs, may be challenging. In recent years, sclerotherapy has become the first-line therapy for most VMs, but surgery still plays a crucial role in select cases. Herein, we report a patient with a large VM in the right lower extremity who achieved favorable outcomes by complete surgical resection.

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Case Report

A 59-year-old man presented to our hospital with a chief complaint of progressive swelling of his right lower extremity associated with pain. He had previously undergone partial resection of a soft-tissue mass in the right thigh during early childhood and his thirties at another hospital. However, the residual mass had gradually increased in size, eventually causing discomfort and pain. The details of the previous surgical treatment were not available. The patient had no other significant medical or family history except laparoscopic cholecystectomy for gallstones 16 years ago. A physical examination revealed a diffuse, compressible swelling extending from the right proximal thigh to the back of the knee (**Fig. 1A**). The swelling was massive, especially in the frontal aspect of the thigh, and a blue discoloration of the skin suggested an invasion of vascular lesions into the skin. No limb-length discrepancy was observed. Laboratory data revealed elevated D-dimer (7.52 μg/mL) and fibrin degradation product (FDP; 15.3 μg/mL) levels. Other laboratory data were within the normal ranges. Short inversion time inversion recovery (STIR) T2-weighted magnetic resonance imaging (MRI) revealed an area of high signal intensity in the subcutaneous tissue of the right lower extremity, indicating a large VM (**Figs. 1B** and **1C**). The lesion was well-circumscribed, and the structures below the muscle fascia were normal. Right femoropopliteal arteriography revealed no evidence of abnormal hypervascular lesions or rapid arteriovenous shunting (**Figs. 1D** and **1E**). Based on these findings, a clinical diagnosis of a slow-flow VM was made. After discussing the case among the relevant departments, complete surgical resection was indicated because the lesion was located in the subcutaneous tissue and was well-circumscribed. The operation was performed under general anesthesia. A skin incision was made over the lesion in a zig-zag manner to prevent scar contracture.

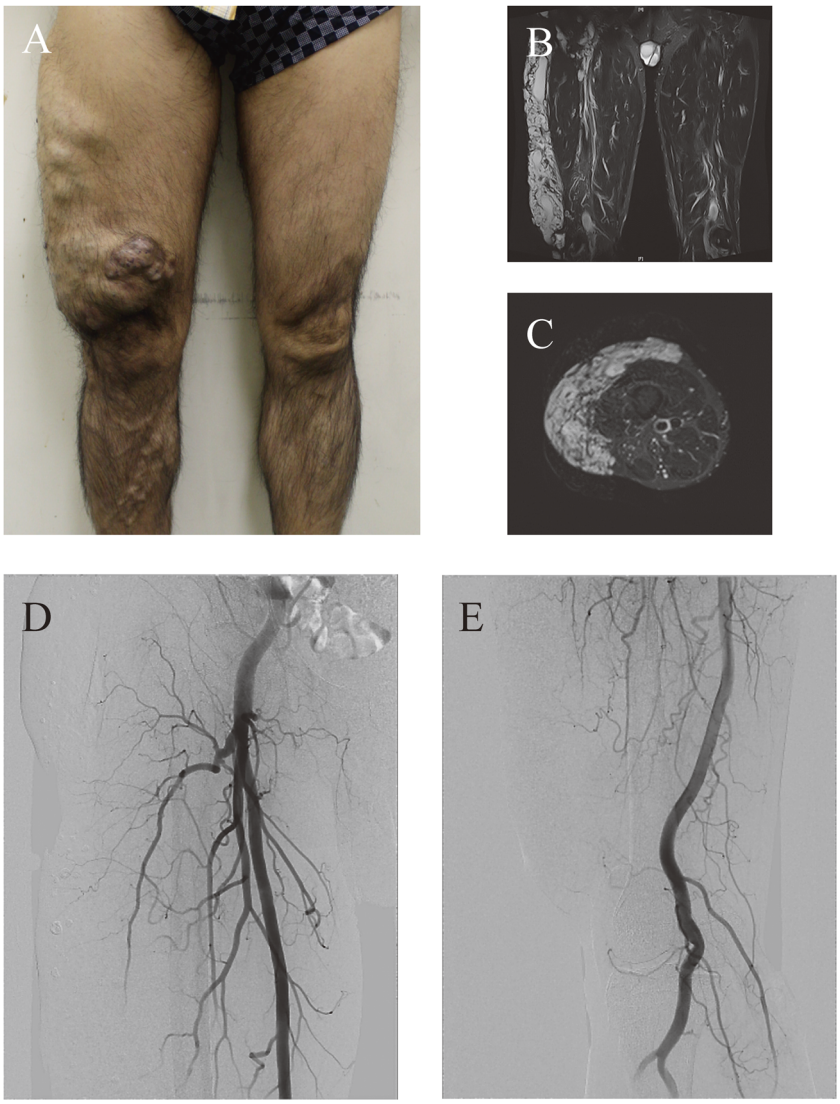

Fig. 1 Preoperative clinical and radiological findings. (**A**) The clinical appearance of a venous malformation in the right lower extremity. (**B**, **C**) Short inversion time inversion recovery (STIR) T2-weighted MRI images showing a large VM with a well-circumscribed margin in the subcutaneous tissue of the right lower extremity. (**D**, **E**) Right femoropopliteal angiography showing no early venous shunting.

The subcutaneous lesion, including the involved skin, was excised. Some perforating veins were ligated, and macroscopic complete resection was achieved. Two subcutaneous drainage tubes were placed, and the skin incision was closed with simple interrupted sutures. A histopathological examination of the specimen revealed dilated capillary and venous channels with sparse smooth muscle cells, which were compatible with the findings of VM (**Fig. 2**). During the postoperative course, minor skin necrosis at the incision line was observed. After wound care with surgical debridement, split-thickness skin grafting was applied on postoperative day 35 for coverage of the wound. No functional impairment associated with the operation was observed. The patient was discharged 52 days after surgery. He was followed up with clinical and ultrasono-

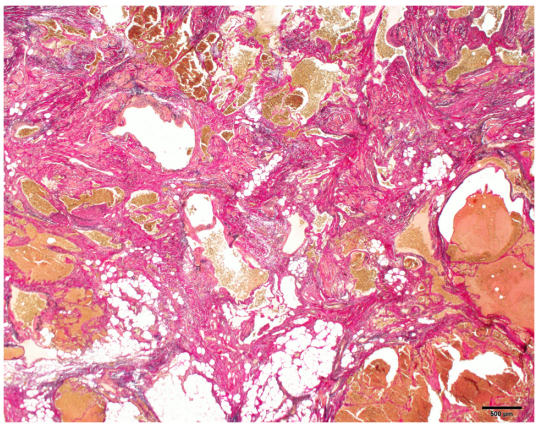

Fig. 2 The histopathological findings show abnormally dilated veins.

Fig. 3 A photograph obtained 8 months postoperatively.

graphic examinations. At 8-month follow up, he was free of symptoms and satisfied with the results (**Fig. 3**).

Discussion

Vascular anomalies are divided into vascular tumors (true neoplasm of endothelial cells) and vascular malformations (structural abnormalities), according to the widely accepted International Society for the Study of Vascular Anomalies classification.[1] This classification was created based on Mulliken and Glowacki's seminal work[2] and revised in 2014. Vascular malformations are further classified based on their channel type (arterial, venous, capillary, lymphatic, or combined) and flow characteristics. VMs are the most common type of vascular malformations, comprising more than 75% of vascular malformations.[3] VMs are typically present at birth and persist and progress throughout life. VMs can affect any body part and different tissue types. The most commonly affected areas are the extremities (40%), the head and neck (40%), and the trunk (20%).[4] VMs in the extremities often present with swelling, pain, bleeding from superficial lesions, deformity, and functional impairments. In the present case, massive swelling and pain indicated the need for treatment. The pain associated with VMs in the extremities can be caused by several factors. Preoperative examinations of our patient revealed no sign of thrombophlebitis or muscle or bone invasion. We considered a massive expansion of the veins to have been the cause of pain. Treatment options for VMs depend on their location and extent. The goal of treatment also depends on the patient's condition. There is no accepted consensus on the treatment of VMs[4] because of the wide variety in their clinical manifestations. Therefore, clinicians often have difficulty in determining the appropriate therapy, especially in cases of large, extensive lesions. The management of VMs is often multistaged and requires a multidisciplinary approach, including interventional radiologists, plastic surgeons, and vascular surgeons. Conservative or compression therapy using an elastic garment can be initially used if the lesions are small and cause minimal symptoms, but most symptomatic patients require further treatment. Sclerotherapy is generally preferred as a minimally invasive treatment method for VMs and can be performed several times. It was reported that sclerotherapy was effective in patients with small VMs, well-circumscribed VMs, and VMs with good stasis of sclerosant.[5] The aim of sclerotherapy is not total cure but size reduction and symptom palliation.

Surgery is now considered as an adjunctive therapy. It is usually performed together with other therapies for lesions that cannot be managed with sclerotherapy alone.[6] Nevertheless, surgery still plays a crucial role in select cases, and the ultimate goal of surgical resection is complete resection to prevent recurrence.[7] In the present case, despite the large, extensive lesion, the decision to perform complete surgical resection was made because the lesion was well-circumscribed on MRI findings. Furthermore, as the lesion was located in the subcutaneous tissue and had not invaded into the muscle or bone, it was unlikely that complete resection would lead to postoperative functional impairment. The MRI findings were the most important information for surgical planning and determining resectability. Considering the size of the lesion and skin involvement, sclerotherapy seemed to be inappropriate in our case. Preoperative angiography was performed to determine if arteriovenous fistulas were present. The major difference between the surgical techniques and concept of high-flow arteriovenous malformations (AVMs) and VMs is the need to effectively control the blood flow. For AVMs, arterial embolization is often used as the main treatment, and preoperative embolization can facilitate surgical resection by reducing intraoperative blood loss.[8] No obvious arteriovenous communications were identified in our patient. Therefore, we proceeded with surgical resection without the need for arterial embolization.

There are some possible drawbacks to radical resection, including non-healing of wounds, injury to adjacent organs, esthetic issues, and the risk of recurrence.[9] The rate of recurrence after the radical resection of VMs is

reported to be 10%. As was observed in our patient, the partial resection of VMs is associated with a higher recurrence rate, and an increase in the size of VMs has been reported in approximately one-fourth of all patients after undergoing debulking surgery.[9] The decision to perform surgery must therefore be made with prudence. Our present patient experienced postoperative skin necrosis, which was successfully managed with local debridement and the application of a split-thickness skin graft. A complete surgical resection of a large VM should be considered only when the expected benefits outweigh the potential risks following the treatment.

Conclusion

Herein, we reported a patient with a large VM in the lower extremity who achieved favorable outcomes by complete surgical resection. Even large VMs can be successfully treated with surgery when patients are properly selected. An accurate and careful evaluation is essential for selecting an appropriate treatment strategy.

Disclosure Statement

All authors have no conflict of interest.

Author Contributions

Study conception: YY, YI
Data collection: YY, KI
Writing: YY
Critical review and revision: all authors
Final approval of the article: all authors
Accountability for all aspects of the work: all authors

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