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An enlarged intramuscular venous malformation in the femoral region successfully treated with complete resection

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ABSTRACT

INTRODUCTION: Intramuscular venous malformations have been previously described as intramuscular hemangiomas, and various therapies have been applied for their treatment. This condition is relatively rare, and therefore, physicians often struggle to determine the appropriate therapy. We presented a case of an enlarged intramuscular venous malformation relapsed after surgery successfully treated with complete resection.

PRESENTATION OF CASE: We presented a case of an enlarged intramuscular venous malformation with postoperative recurrence successfully treated with complete resection. A 63-year-old woman presented with a subcutaneous mass in the right distal thigh. She experienced swelling in the right thigh 19 years previously and was diagnosed with a venous aneurysm. Three-dimensional CT angiography confirmed the presence of an irregular vessel assumed to be the feeding vessel, which was dendritically branched from the deep femoral artery. We performed surgical complete resection. Her pain and gait disturbance improved after surgery, and she has not experienced recurrence of the mass for the past 2 years.

DISCUSSION: Conservative therapy is initially used for venous malformations. Sclerotherapy, laser therapy, or surgical resection is considered after low-dose aspirin therapy, in combination with the use of compressive garments. Surgical resection is indicated for completely resectable lesions and is appropriate for large lesions in terms of cosmetic benefit. However, partial resection may result in excessive bleeding or postoperative recurrence.

CONCLUSION: The therapy for venous malformations should be decided based on the degree of disability in daily living, adjacent tissue damage, and cosmetic concerns after appropriate differential diagnostic investigations and biopsy.

1. Introduction[1]

Vascular lesions classification system and their detailed division into groups and subgroups were elaborated and implemented in Rome, in 1996, during the International Society for the Study of Vascular Anomalies (ISSVA) [1]. Prior to the classification is established, intramuscular venous malformations have been previously described as intramuscular hemangiomas, and various therapies have been applied for their treatment. This condition represent 0.8% of all hemangiomas. The most common location is the lower extremities (45%), followed by the upper extremities (27%) and head and neck (14%). They tend to appear in adolescence or young adulthood, and can cause pain and swelling that worsens during physical activity [2]. The therapeutic method depends on the type and extensiveness of the malformation, its clinical symptoms and patient's age [3]. Therefore, physicians often struggle to determine the appropriate therapy. Here we present a case of an enlarged intramuscular venous malformation in the femoral region successfully treated with complete resection.

2. Presentation of case

A 63-year-old woman presented with a subcutaneous mass in the right distal thigh. She had no relevant family history. However, she had undergone surgical resection of a hemangioma in the right thigh 19 years previously. She experienced swelling in the right thigh 19 years previously and was diagnosed with a venous aneurysm. She therefore underwent surgery; however, a walnut-sized subcutaneous mass remained in the right thigh after surgery, which gradually enlarged. She was kept under observation as no subjective symptoms were noted; however, she eventually developed gait disturbance due to the weight of the enlarged mass. She visited a local doctor who referred her to our department.

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[1] ISSVA: International Society for the Study of Vascular Anomalies.

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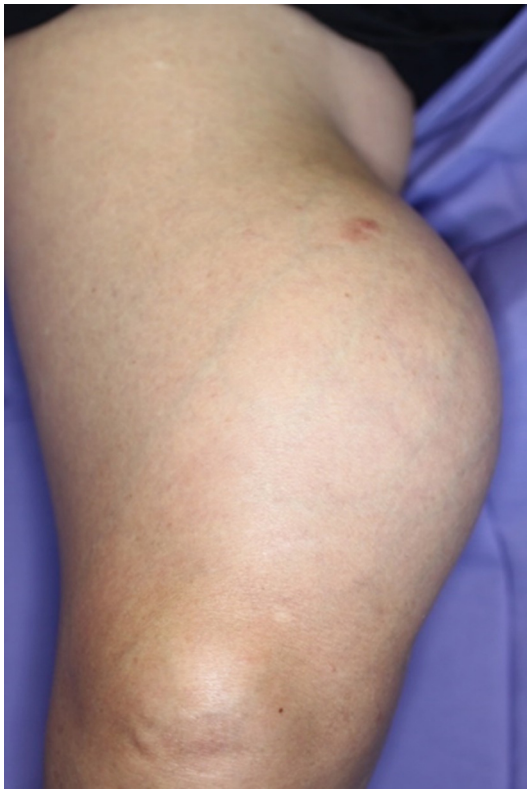

Fig. 1. Clinical findings at the initial visit. A 20 × 15 cm subcutaneous mass with tenderness is noted at the medial side of the right thigh.

A large mass (20 × 15 cm) was noted at the medial side of the right distal thigh. Palpation revealed that its center was soft and its margin was hard and that it was attached to the fascia. Additionally, mild tenderness was observed (Fig. 1).

T2-weighted magnetic resonance imaging (SIEMENS MAGNETOM Symphony QUANTUM®) indicated an intramuscular solid mass with an irregular intensity structure (Fig. 2a). Computed tomography (CT: SIEMENS SOMATOM Definition Flash®) showed that the mass had a high density, similar to that of a muscle. Enlarged vessels were identified around the mass, and contrast-enhanced CT showed slow contrast enhancement mainly at the margin of the mass. Three-dimensional CT angiography confirmed the presence of an irregular vessel assumed to be the feeding vessel, which was dendritically branched from the deep femoral artery (Fig. 2b). A vascular malformation was suspected because the mass developed over a long period (19 years), and invasive expansion to the surrounding tissues was not observed despite the expansive growth pattern of the mass on imaging examinations. However, an irregular internal structure was observed. Thus, we attempted to exclude the possibility of soft-tissue sarcoma and definitively diagnose the mass after incisional biopsy.

In the biopsy samples, striated muscle tissue was observed with vascular proliferation at the margins. Vessels with various diameters were observed; however, no apparent nuclear atypia or nuclear division was noted. Hence, she was considered to have a venous malformation.

She experienced pain and gait difficulty. Additionally, her activities of daily living (ADL) were affected. Thus, surgical complete resection was performed. Intraoperatively, the mass capsule was relatively clear; thus, resection was initiated from the capsule margin, and the femoral artery was identified in the deep area, which was assumed to be the feeding vessel (Fig. 3). The bifurcation of the vessel supplying the mass, which was a branch of the femoral artery, was ligated, and then the mass was removed. A part of the mass was in contact with the femur with mild adhesion. However, the femur was deformed owing to the expansive growth of the mass.

The mass was covered with a capsule-like fibrous stroma and had a dense fibrous stroma and cavernous enlarged vascular lumen filled with erythrocytes (Fig. 4a and b). A magnified image indicated no nuclear atypia and very few nuclear divisions in the vascular endothelial cells (Fig. 4c). Immunohistochemical staining showed that CD31, CD34, and α-SMA (smooth muscle) were positive and AE1/AE3 and HHB-8 were negative in the endothelial cells. Based on these findings, she was diagnosed with an intramuscular venous malformation.

Her pain and gait disturbance improved after surgery, and she has not experienced recurrence of the mass for the past 2 years (Fig. 5).

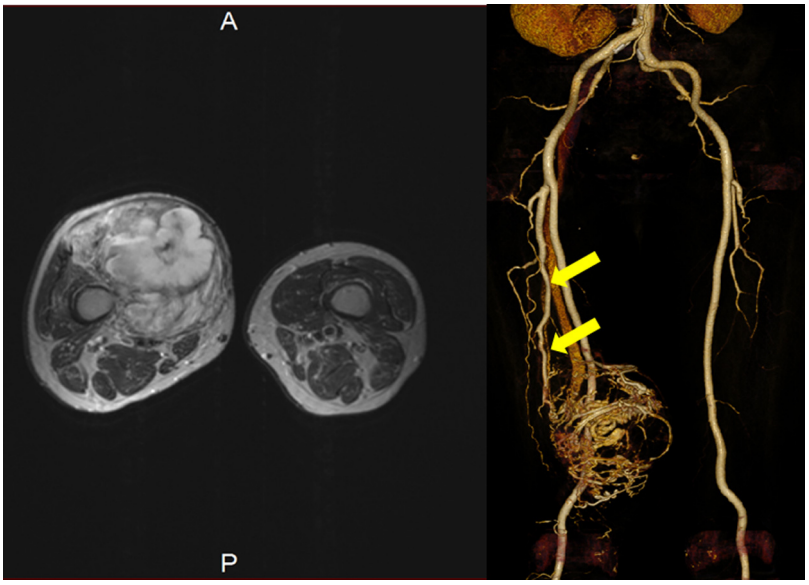

Fig. 2. Magnetic resonance imaging (MRI) and computed tomography angiography (CTA) findings. (a) An axial T2-weighted MRI image showing a solid mass with a high intensity signal, (b) a 3-dimensional CTA image showing an irregular vessel assumed to be the feeding vessel. The arrows were dendritically branched from the deep femoral artery.