



Intramuscular Arteriovenous Malformation of the Gluteus Maximus Muscle

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Extracranial arteriovenous malformations (AVMs) are far less common than intracranial AVMs. AVMs mostly involve the skin, subcutaneous tissue and even the musculoskeletal system extensively. The goal of treatment is to control the AVM, using tumor debulking or arterial embolization; complete tumor excision is usually not possible and the local recurrence rate is high. Intramuscular AVMs are rare and these lesions can be curable with complete surgical resection. We present a case of a rare intramuscular AVM of the right gluteus maximus muscle in a 33-year-old man. The AVM had undergone progressive enlargement and produced pulsation and pain. We performed selective embolization and radical excision of the AVM with removal of the entire muscle. There was no local recurrence of the AVM and the patient's right hip extension and external rotation had recovered well at a 12-month follow-up. AVMs are extremely rare in skeletal muscle and less than 1% of vasoformative tumors throughout the body occur in such locations. The treatment of choice is a combination of selective embolization and radical excision to avoid recurrence. In fact, intramuscular AVMs are curable compared with those in other sites.

Key words: arteriovenous malformation (AVM); intramuscular AVM

INTRODUCTION

Arteriovenous malformations (AVMs) are high-flow lesions providing a direct connection between an artery and a vein. AVMs are usually latent during infancy and childhood and may enter an active expanding phase in adolescence. Extracranial AVMs are far less common than intracranial AVMs.¹ These lesions often occur on the extremities or trunk where they may present as an enlarging soft tissue mass in the subcutaneous tissue, or may be located below the deep fascia and involve the musculoskeletal system. Less than 1% of vasoformative tumors throughout the body occur in the skeletal muscle and AVMs here are extremely rare.² Although the mechanisms are poorly understood, infection, trauma, or

hormonal changes in puberty and pregnancy are known factors that can trigger the rapid expansion of AVMs. Schobinger described a useful clinical staging system that describes their progression.¹ AVMs initially present as warm pink-blue macules (stage I), proceed to enlarge with pulsations, thrills and bruits (stage II), subsequently can become painful, bleed or ulcerate (stage III) and finally can result in cardiac failure (stage IV).

Magnetic resonance imaging (MRI) is helpful in determining the extent of the lesion and any involvement of associated structures. Angiography is the ideal modality to determine the anatomy of the feeding and draining vessels including the extent of arteriovenous shunting, fistulae and vessel tortuosity. Angiography can also be used for therapeutic embolization of the AVM.⁴ Usually, surgical management is difficult because of the major blood loss that can occur leading to incomplete removal of the lesion and its recurrence. Excessive bleeding during operation can be prevented by preoperative embolization of the AVM. Conservative treatment is often suggested in asymptomatic cases because of the high recurrence rate after treatment and difficulties during operation. AVMs mostly involve the skin, subcutaneous tissue and even the musculoskeletal system extensively. The goal of treatment is to control the AVM using tumor debulking or arterial embolization; complete tumor exci-

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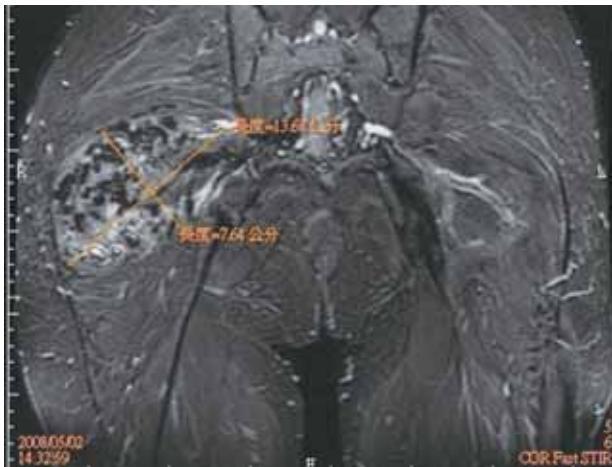


Fig. 1 A large mass is shown within the gluteus maximus muscle with a high signal on T2-weighted magnetic resonance imaging (MRI) indicating enlarged venous compartments. The greatest dimension was more than 13 cm. Tubular structures with low signals indicate fast voiding vessels.

sion is usually not possible and the local recurrence rate is high. In contrast, intramuscular AVMs can be curable with complete surgical resection. Intramuscular AVMs have been reported in the piriformis muscle, flexor digitorum superficialis and in the temporoparietal, temporal and lingual musculature.⁵⁻⁹ According to review articles, this is the first reported case in the English literature of an intramuscular AVM of the gluteus maximus muscle.

MATERIALS AND METHODS

A 33-year-old man presented with a large mass in his right gluteus maximus muscle. The mass had undergone rapid and painful enlargement with pulsation over the previous months. MRI showed a large soft tissue mass with a low signal on T1-weighted images and a high signal on T2-weighted images, indicating enlarged venous compartments (Fig. 1). The diagnosis of AVM was confirmed by intra-arterial angiography, which showed a nidus type lesion measuring 81×86 mm with feeder vessels arising mainly from the branches of the right internal iliac artery (superior and inferior gluteal arteries). To avoid excessive intraoperative bleeding, we performed selective embolization before operation and radical excision of the AVM with removal of the entire gluteus maximus muscle three days later. Embolization of the malformation using four bottles of 33% n-butyl-cyanoacrylate glue through superselective microcatheters was

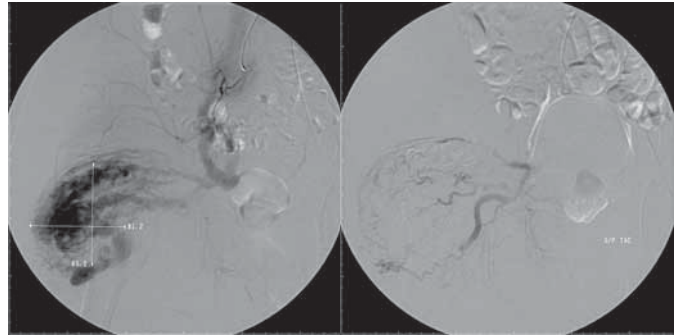


Fig. 2 Left: angiography showed a nidus type lesion measuring 81×86 mm with feeder vessels arising mainly from branches of the right internal iliac artery. Right: after selective embolization of the feeding arteries, the residual blood flow was reduced markedly.

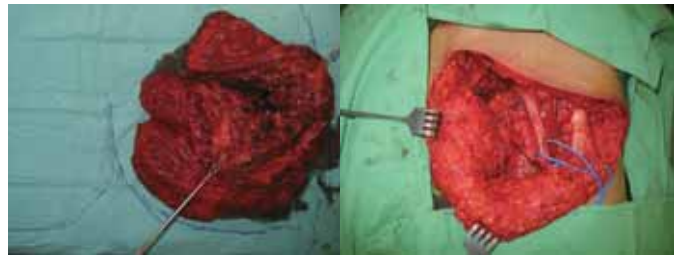


Fig. 3 Left: the right gluteus maximus muscle, including the AVM measuring about 12×7 cm, was excised entirely. Multiple engorged, dilated and haphazardly distributed large vessels were noted within the muscle. Right: the right sciatic nerve was preserved intact.

performed via the distal branches of the right internal iliac artery. Five fibered coils were disposed into these branches. After embolization, the residual blood flow was reduced markedly (Fig. 2). Surgical resection was done within 72 hours. The AVM was entirely within the gluteus maximus muscle and multiple engorged, dilated and haphazardly distributed large vessels were noted. The lesion measured about 12×7 cm (Fig. 3). The pathology results demonstrated thickening of the venous intima (arterialization) and tortuosity of the muscular vessels, indicating an intramuscular AVM (Fig. 4). There was no local recurrence of the AVM at a 12-month follow-up. The right sciatic nerve was preserved intact after removal of the overlying muscle (Fig. 3). De-epithelialization of the inferior skin flap surrounding the incisional wound was performed appropriately to increase soft tissue thickness beneath the upper skin flap and to protect the sciatic nerve from external compression. This also served to improve the contour of the right buttock.

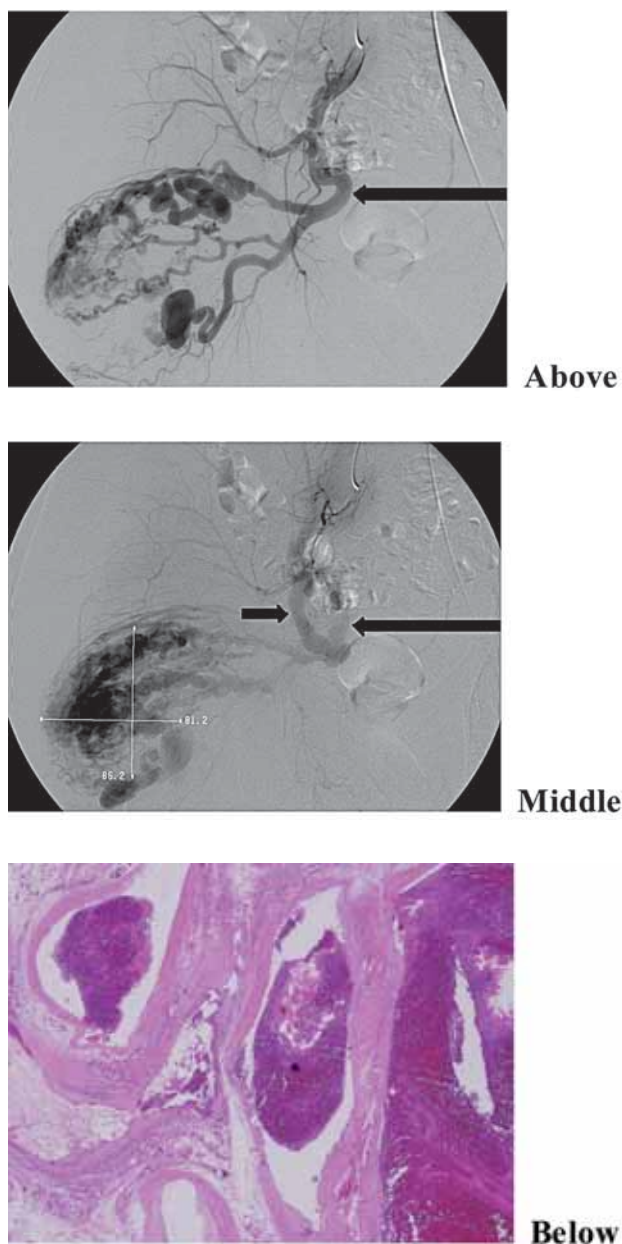


Fig. 4 **Above:** angiography showed a large AVM with feeding arteries arising from the distal branches of the superior and inferior gluteal arteries (long arrow). **Middle:** the feeding artery (long arrow) and the early draining vein (short arrow) were both found at the same view of angiography and it confirmed the property of high flow AVM. **Below:** microscopically, the sections show a picture of multiple haphazardly distributed large muscular vessels and thickening of the venous intima (arterialization), consistent with intramuscular AVM according to angiography which reveals feeding arteries and drainage veins. (H&E, original magnification $\times 40$)

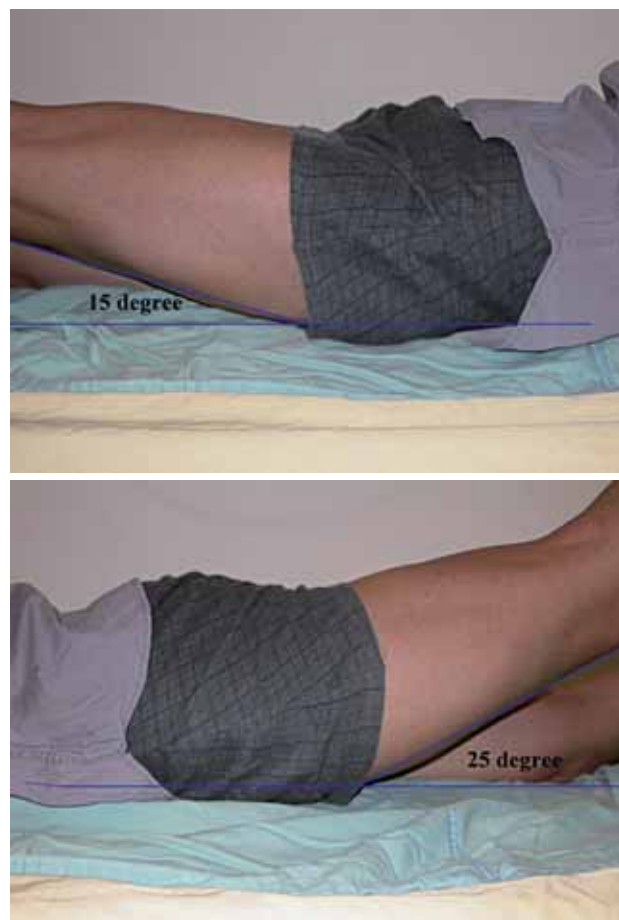


Fig. 5 When one is examining the extension of the hip, the patient needs to be instructed to extend the thigh posteriorly while prone.¹⁴ The patient has 15 degrees at the right hip(affected side) (photography, above) and 25 degrees at the left hip(photography, below) respectively in the hyperextension position. Extension of the right hip was almost normal at the 6-month follow-up.

RESULTS

The patient reported sensory disturbances of his right buttock and walking with a cane was necessary during the first month postoperatively because of weakness in the right hip. After a laborious rehabilitation, the patient can now walk independently for daily activities without assistance. The patient's right hip extension, external rotation and ability to arise from a sitting position recovered well after training muscles in the right thigh to compensate for the function of the gluteus maximus muscle (Fig. 5). MRI imaging at a 4-month follow-up confirmed complete excision of the AVM and no surgery-related

complications or obvious morbidity were detected at a 12-month follow-up.

DISCUSSION

Hemangiomas and vascular malformations are benign lesions of blood vessels. To date, many different terms and classifications have been suggested for these types of lesions. Today, most reports use the classification scheme suggested by Mulliken and Glowacki in 1982, based on the histological and clinical features of the lesion. This system divides vascular anomalies into hemangiomas, which are neoplastic lesions with endothelial hyperplasia, and vascular malformations, which are congenital lesions with normal endothelial turnover.¹⁰ The prevalence of vascular malformations is estimated to be 1.5% in the general population, most commonly involving the head and neck, followed by the lower extremities and trunk.¹¹ AVMs mostly involve the skin and intramuscular AVMs are rare. There is no reported gender difference in incidence. Because intramuscular AVMs are rare, to our knowledge, there are no reported data on their exact incidence. The natural history of these lesions follows four stages as described by Schobinger: quiescence, expansion, destruction and decompensation.¹ Clinical examinations and patient history are usually adequate for an accurate diagnosis of these lesions. Plain or color Doppler ultrasonography is effective in evaluating these malformations, which appear as heterogeneous lesions with large vessels and multiple sites of pulsatile arteriovenous shunting. MRI or angiography is an excellent modality for evaluating the local extent of arteriovenous malformations and for separating them from other venous malformations. Angiography is helpful for those patients requiring surgical intervention or preoperative embolization.^{10,12} AVMs can prove deceptively problematic and even dangerous to treat. Thus, most authors agree that conservative treatment is preferable in the absence of significant symptoms.¹⁰⁻¹² When surgery is required, angiographic studies followed by selective embolization is usually done 24-72 hours before resection. It should be emphasized that embolization or ligation of the feeding arteries without surgical resection is strongly contraindicated because occlusion of these arteries usually results in rapid recruitment and dilation of previously microscopic collateral blood vessels.^{11,12}

In our case, the patient underwent preoperative angiography for the evaluation and selective embolization of feeding arteries that could help to reduce intraoperative bleeding. The diagnosis of AVM was confirmed

by intra-arterial angiography, which showed feeding arteries arising from the distal branches of the superior and inferior gluteal arteries and an abnormal vascular network (Fig.2).⁸ Three days later, surgical resection was performed with the patient under general anesthesia in a prone position. Because of the properties of intramuscular lesions, the AVM in this case was permanently curable by complete excision of the lesion in the gluteus maximus muscle after ligation of feeding arteries (superior and inferior arteries). The sciatic nerve was preserved intact and this reduced postoperative morbidities. The gluteus maximus muscle extends the thigh at the hip, assists in its lateral rotation, steadies the thigh and assists in raising the trunk from a sitting position. The normal range of motion at the hip in the hyperextension position, the main action of the gluteus maximus, is 10-30 degrees. After resection of gluteus maximus muscle, hip extension is compensated gradually by thigh extension using the semitendinosus, semimembranosus and biceps femoris muscles and muscles such as the piriformis, obturator externus and quadratus femoris will compensate to enable external hip rotation.¹³ The patient has 15 degrees of motion at the right hip (affected side) and 25 degrees at the left hip in the hyperextension position (Fig. 5). The hip's motion had recovered after proper rehabilitation without obvious morbidity.

CONCLUSION

Because it is dangerous to treat AVMs with surgical intervention that may cause excessive blood loss, many authors suggest conservative treatment for asymptomatic cases.¹⁰⁻¹² Nevertheless, therapies for intramuscular AVMs may not be the same as for other AVMs. Intramuscular AVMs may be curable by complete excision of the muscle and by preserving adjacent vital structures that will help to reduce postoperative morbidity. To achieve a permanent cure, a combination of surgery and preoperative selective embolization to reduce intraoperative bleeding can be the best choice in the treatment of resectable AVMs such as intramuscular ones.

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