Coil embolization of a gluteal false aneurysm in a patient with Marfan syndrome

M. A. Vasseur, MD, V. C. Doisy, MD, A. G. Prat, MD, and **C. Stankowiak, MD,** *Lille, France*

Gluteal aneurysms, whether true or false, are exceptional. They represent less than 1% of all aneurysms and develop within the superior or inferior gluteal arteries, being branches of the internal iliac artery. We report here the case of a 35-year-old patient with Marfan syndrome in whom annuloaortic ectasia and Barlow's disease with mitral valve insufficiency successively developed followed by a gluteal false aneurysm, which led us to investigate the etiologic mechanism of the patient's conditions. The gluteal aneurysm was successfully treated by selective embolization, which would appear to be the elective therapeutic approach for these lesions. (J Vasc Surg 1998;27:177-9.)

The classic cardiovascular complications of Marfan syndrome are annuloaortic ectasia, mitral valve disease, and aortic dissection. These conditions can be explained by a progressive loss of connective tissue elastic fibers of the inner arterial wall that lead to its fragility and distensibility. Other aneurysmal locations have been reported in relation to this condition, though clearly less frequently. We report here the case of a patient with Marfan syndrome who successively had annuloaortic ectasia and Barlow's disease, which were successfully treated in 1987, a false aneurysm on the reimplantation of the right coronary artery that necessitated reintervention in 1992, and a gluteal false aneurysm diagnosed and treated in 1993.

CASE REPORT

A 35-year-old patient (height, 202 cm; weight, 66 kg) with a history of recurring pneumothorax treated by pleurotomy, familial lens luxation, and Barlow's disease, underwent operation in 1987 for annuloaortic ectasia associated with a mitral insufficiency. The patient underwent a composite graft replacement of aortic valve and ascending aorta with reimplantation of both coronary arteries associated to a quadrangular resection of the posterior mitral leaflet. Medical follow-up was uneventful and consisted of an annual duplex scan and thoracic contrast-enhanced computed tomographic (CT) scan, which were unremarkable until 1992, when a voluminous false aneurysm at the level of the right coronary artery (RCA) reimplantation was discovered. The right coronary artery was thrombosed. Reintervention revealed a complete dehiscence of the anastomosis. The prosthetic rupture was closed by running suture, and the thrombosed nonreimplantable right coronary artery was ligated. After this procedure, the patient did well.

Eighteen months later, the patient noticed the appearance of a painless, nonpulsatile mass in the left gluteus. Intramuscular premedication had been administered in the left buttock before all these operations. Ultrasound revealed a fluid-filled mass, and a duplex scan detected pulsatile flow within it. An arteriogram confirmed the diagnosis of a false aneurysm of the left superior gluteal artery (Fig. 1). Selective percutaneous embolization via the right femoral artery was performed, using an 8F catheter to place five coils (diameter, 2 mm) within the aneurysm, and its principle afferent, an accessory branch, was occluded by a 3 mm diameter coil using a 0.25F microcatheter. Thrombosis of the false aneurysm was immediately achieved (Fig. 2); a duplex scan and angiogram 3 months later confirmed this outcome. The patient has been followed-up for 36 months and continues to do well.

DISCUSSION

Marfan syndrome is a hereditary autosomal dominant disorder of variable expression and penetrance involving connective tissue and is related to mutations linked to genes coding for fibrillin, the principal component of elastin microfibrils. Kainulainen et al.,[1]

From the Department of Cardiovascular Surgery, Cardiologic Hospital.
Reprint requests: M. A. Vasseur, MD, Department of Cardiovascular Surgery, Cardiologic Hospital, 59037 Lille Cedex, France.

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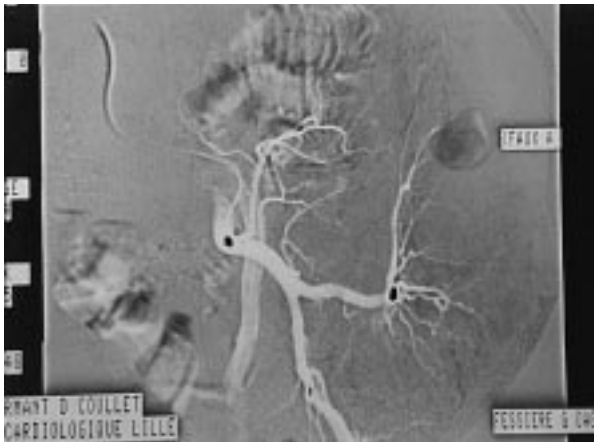

Fig. 1. Gluteal false aneurysm.

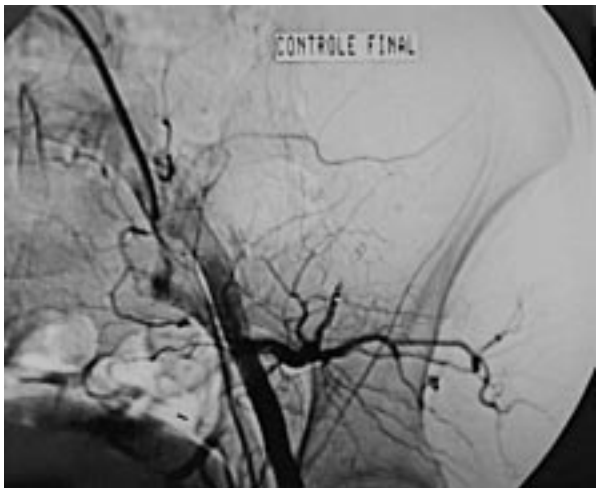

Fig. 2. Control after embolization.

using the linkage approach, succeeded in establishing a "Marfan locus" on chromosome 15. The typical clinical manifestations in the eyes, skeleton, and cardiovascular system appear to relate directly to weakening of the supportive tissues.

The most frequently observed cardiovascular anomalies are mitral and aortic valve disease,uloaortic ectasia, and aneurysms of the ascending thoracic aorta and aortic dissections.[2] More rarely reported are cases of aneurysms of the brachiocephalic arteries, descending thoracic aorta, or infrarenal aorta. The pulmonary artery has also been involved with an aneurysmal dilatation.[3] Aneurysmal involvement of peripheral arteries is rare, and it does not seem that Marfan syndrome predisposes them to this. We therefore hypothesized that the gluteal aneurysm of our patient was iatrogenic and related to intramuscular injections in the left buttock, as already described by Vauthey et al.[4]

Most gluteal false or true aneurysms, which represent less than 1% of all aneurysms,[5] result from blunt or penetrating injuries. Usually pelvic fractures damage the inferior gluteal artery or its branches, whereas penetrating traumas affect the superior gluteal artery.[6] The delay between trauma and diagnosis can vary from a few weeks to several years. Other etiologic mechanisms that have been described are periarteritis nodosa, atheromatosis, mycotic aneurysms, arterial dysplasias, and aneurysmal evolution of persistent sciatic artery.[7]

The clinical expression of gluteal aneurysms is variable. Manifestations include a pulsatile painful buttock mass, rupture, and when large, compartment syndrome or compression of the sciatic nerve.[8] Pulsatility of these aneurysms is inconstant, explaining reported errors in diagnosis. Sometimes, such an aneurysm may be mistaken for a soft tissue tumor[9] or an abscess because of cutaneous erythema or an inflamed appearance.

Duplex scanning allows confirmation of the arterial origin of the mass, whereas the size and topographic relations of aneurysms can better be defined by CT scan or nuclear magnetic resonance examination.[10] Arteriography is indispensable, permitting clarification by selective injection, if needed, of the artery or branches involved in the aneurysmal process.

The treatment of these lesions has been, for a long time, exclusively surgical. The direct approach, occasionally carried out when the diagnosis of an abscess was made, is not recommended. Dissection through the gluteus maximus and gluteus medius is difficult and carries the risk of trauma to the sciatic nerve, muscular necrosis, and, above all, problems to control major hemorrhage. Several authors have therefore proposed preliminary ligation of the internal iliac artery via a retroperitoneal approach or control by an inflated balloon. Burchell[11] has demonstrated that ligation of the internal iliac artery only resulted in a 50% decrease in efferent flow because of collateral pathways between the gluteal, lumbar, sacral, and anorectal arteries. This technique is therefore inappropriate in the treatment of gluteal aneurysms.

The failure of internal iliac artery ligation led to the use of percutaneous embolization as the preferred approach to serious hemorrhage after pelvic trauma, and later as an effective solution to treat gluteal aneurysms.[12] This technique requires selective catheterization of the internal iliac artery and

identification of all afferent and efferent branches of the aneurysm. Indeed, embolization itself is insufficient; all entries and exits must also be selectively or superselectively embolized to achieve complete thrombosis of the aneurysm. Different materials can be used for this purpose; coils of variable diameters and inflatable balloons are the most commonly used. Thrombosis of the aneurysm is achieved in the majority, although in the case of large aneurysms this sometimes proves impossible to achieve. In the latter, preoperative embolization considerably decreases the risk of subsequent surgical treatment.

CONCLUSION

Even though rare, gluteal true or false aneurysms must be the principal concern in dealing with all gluteal masses. Diagnosis is confirmed by duplex scan and arteriogram, but CT scanning and nuclear magnetic resonance imaging are useful for the clarification of the size and topography of the aneurysm. In the modern era, the preferred treatment of gluteal aneurysms is selective percutaneous embolization, which usually results in definitive control.

REFERENCES

1. Kainulainen K, Pulkkinen L, Savolainen A, Kaitila I, Peltonen L. Location on chromosome 15 of the gene defect causing Marfan syndrome. N Engl J Med 1990;323:935-9.
2. Gery JL Jr, Morris L, Pyeritz RE. Clinical management of the cardiovascular complications of the Marfan syndrome. J La State Med Soc 1991;143:43-51.
3. Disler LJ, Manga P, Barlow JB. Pulmonary arterial aneurysms in Marfan syndrome. Int J Cardiol 1988;21:79-82.
4. Vauthey JN, Maddern GJ, Balsiger D, Blumgart LH, Triller J. Superselective embolization of superior gluteal artery pseudoaneurysm following intramuscular injection: case report. J Trauma 1991;31:1174-5.
5. Demetriades D, Rabinowitz B, Sofianost C. Gluteal artery aneurysms. Br J Surg 1988;75:494.
6. Gilroy D, Saadia R, Hide G, Demetriades D. Penetrating injury to the gluteal region. J Trauma 1992;32:294-7.
7. Natali J, Denis PJ, Kieffer E, Merland JJ. Syndrome de la fesse battante. J Mal Vasc 1989;14;3:183-9.
8. Schmalzried TP, Eckardt JJ. Spontaneous gluteal artery rupture resulting in compartment syndrome and sciatic neuropathy. Clin Orthop 1992;275:253-7.
9. Simon MA, Sailly RE, Springfield DS. Case record of the Massachusetts General Hospital. Case 32. N Engl J Med 1992;327:412.
10. Zirinsky K, Markisz JA, Auth YH, Rubenstein WA, Rozenblit G, Kazam E. Computed tomography and magnetic resonance imaging of a superior gluteal artery pseudoaneurysm. J Comput Tomogr 1988;12:75-8.
11. Burchell RC. Physiology of internal iliac artery ligation. J Obstet Gynecol Brit Comm 1968;75:642-51.
12. Grand C, Delcour C, Bank WO, Braude P, Dereume JP, Struyven J. Emergency embolization of a mycotic aneurysm of the superior gluteal artery: case report. Cardiovasc Intervent Radiol 1992;15:117-9.

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