Successful treatment of a ruptured profunda femoris artery aneurysm in association with fibromuscular dysplasia

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ABSTRACT
Fibromuscular dysplasia is rarely reported in the infra-inguinal arteries. We report a case of an 86-year-old woman who presented with a ruptured profunda femoris artery aneurysm who was found to have angiographic findings of fibromuscular dysplasia in the bilateral deep femoral arteries and bilateral renal arteries. The rupture was treated successfully with a balloon-expandable covered stent. (J Vasc Surg Cases and Innovative Techniques 2019;5:485-7.)

Keywords: Fibromuscular dysplasia; Aneurysm; Profunda femoris; Rupture; Endovascular

CASE REPORT
An 86-year-old nonsmoking woman with a past medical history significant for atrial fibrillation on warfarin, a permanent pacemaker, hypertension on a beta-blocker and calcium channel blocker, and dyslipidemia was transferred to our institution for a large left thigh expanding hematoma and anemia. The patient denied any recent history of trauma.

On arrival to the emergency room she had a heart rate of 110 beats per minute with a blood pressure of 102/63 mm Hg. Physical examination revealed palpable bilateral femoral pulses and pedal signals present with Doppler bilaterally. Her left thigh had a tender, nonpulsatile mass approximately 5 × 10 cm at the skin level, which was not actively expanding.

Diagnostic studies included a complete blood count and basic metabolic panel as well as a prothrombin time, international normalized ratio, and a partial thromboplastin time. Her hemoglobin was 6.5 g/dL, for which she was transfused 2 units of packed red blood cells. Her international normalized ratio was 3.2, for which she was transfused 2 units of fresh frozen plasma.

Computed tomographic angiography demonstrated a complex multifocal, saccular aneurysm involving the proximal profunda femoris artery (PFA), measuring 3.9 × 2.6 cm with peripheral calcification and a large hematoma measuring 10 × 14.4 × 13.6 cm within the anterior compartment of the thigh with active extravasation (Fig 1). She had no family history of aneurysmal disease, collagen vascular disorders, or renal artery disease.

The patient was taken to the hybrid operating room. Access angiogram on right showed aneurysmal dilatation of the right PFA with a beads on a string appearance (Fig 2). An aortogram with bilateral pelvic runoff demonstrated the same beads on a string of the renal arteries, left greater than right (Fig 3). Selective angiogram of the left leg demonstrated active extravasation of contrast from the left PFA (Fig 4). The left PFA also had the same beads on a string appearance. After the common femoral artery and the origin of the PFA were marked, a covered-balloon-expandable stent (VBX, W. L. Gore & Associates, Flagstaff, Ariz) was deployed, covering the deep femoral artery ostium. A completion angiogram of the left lower extremity demonstrated resolution of extravasation from the left PFA (Fig 5).

Her postoperative recovery was complicated by uncontrolled atrial fibrillation with a rapid ventricular rate, controlled with an increase in her metoprolol dose and the addition of digoxin. She required no further blood transfusions. She was restarted on her warfarin postoperative day 4 and was discharged to rehabilitation on postoperative day 9. In addition, bilateral carotid duplex examination revealed only mild disease without any evidence of fibromuscular dysplasia (FMD). On her postoperative follow-up visit, the patient was doing well and completing rehabilitation. She had no evidence of bleeding with normal vital signs and the hematoma was decreased in size. The patient was able to ambulate with assistance and denied any symptoms of claudication or rest pain. Patient publication consent was obtained at this time.

DISCUSSION
We report a case of multianeurysmal degeneration of the PFA in association with other lesions suggesting FMD and presenting as a rupture.

PFA aneurysm is rare, accounting for only 0.5% of all peripheral aneurysms. They are difficult to diagnose in the absence of complications or symptoms owing to their location in the deep thigh musculature. As a result, rupture is the most common presentation, occurring in 13% to 45% of cases. Previous studies have indicated that patients often have other aneurysms in up to 75% of cases, such as in the abdominal aorta or popliteal artery. Although the literature is scarce, demographic
information regarding PFA aneurysm indicate that the mean age is in the mid-70s, and that males are predominantly affected.4

Our angiographic findings of a beads on a string appearance of bilateral profunda femoris arteries and bilateral renal arteries were most suggestive of FMD (Figs 2 and 3). FMD is a nonatherosclerotic, noninflammatory vascular disease that most commonly involves the carotid, renal and other medium-sized vascular beds with few primary branches. Uncommon locations of FMD include the brachial artery and external iliac artery.5-9 However, this pathology rarely involves the infra-inguinal vascular distribution, such as the PFA. In prior reports of infra-inguinal FMD in the literature, the presenting symptoms included intermittent claudication, critical limb ischemia, or peripheral microembolism.
none of which have been described as presenting as a rupture.\textsuperscript{5-9} Iwai et al\textsuperscript{10} reported three cases of infra-inguinal arterial FMD, one of which involved the PFA. Schneider et al\textsuperscript{11} reported a case of a Caucasian woman with FMD of the PFA presenting as isolated thigh claudication, treated with surgical bypass of the FMD segment. Additionally, Esfahani et al\textsuperscript{12} reported a case series of 17 Iranian men with FMD of the lower extremities who sought treatment for lower extremity ischemia symptoms. Only four of these cases involved the PFA.

Treatment options for this case included both an endovascular and an open option. In addition to the endovascular approach described, coil embolization of the PFA before exclusion with a covered stent would have also been an acceptable option. An open repair with exclusion of the ruptured aneurysm and evacuation of hematoma would have been an acceptable open option. In our case specifically, we decided to treat by exclusion with a balloon-expandable covered stent and, if the hematoma continued to expand after exclusion of the PFA, we planned to treat percutaneously. The PFA would be easily accessible percutaneously and it could be coil embolized with the addition of thrombin injection if needed. A second option would be an open procedure to gain control and exclude the ruptured aneurysm.

The recommended treatment for FMD in general is for a balloon angioplasty of the effected artery and only if symptomatic.

Although, the diagnosis of FMD requires a tissue diagnosis, our images are highly suggestive of a dysplasia component. This case illustrates the heterogeneous nature of the demographic, anatomic, and clinical presentations of PFA aneurysms and to our knowledge, this is the first reported case of such aneurysms with a beads on a string appearance, suggestive of FMD.

Further investigation is warranted regarding this complicated disease process as well as the efficacy of available treatment options.

**CONCLUSIONS**

We report a rare case of multianeurysmal degeneration of bilateral profunda femoris arteries in association with other lesions suggesting FMD. Treatment consisted of successful deployment of a covered balloon-expandable stent.

**REFERENCES**

1. Harbuzariu C, Duncan A, Bower T, Kalra M, Gloviczki P. Profunda femoris artery aneurysms: association with aneurysmal disease and limb ischemia. J Vasc Surg 2008;47:31-4.
2. Tait W, Vohra R, Carr H, Thomson GJ, Walker MG. True profunda femoris aneurysms: are they more dangerous than other atherosclerotic aneurysms of the femoropopliteal segment? Ann Vasc Surg 1991;5:92-5.
3. Johnson C, Goff J, Rehrig S. Asymptomatic profunda femoris artery aneurysm: diagnosis and rationale for management. Eur J Vasc Endovasc Surg 2002;24:91-2.
4. Posner S, Wilensky J, Dimick J, Henke PK. A true aneurysm of the profunda femoris artery: a case report and review of the English language literature. Ann Vasc Surg 2004;18:740-6.
5. Lin WW, McGee GS, Patterson BK, Yao JST, Pearce WH. Fibromuscular dysplasia of the brachial artery: a case report and review of the literature. J Vasc Surg 1992;16:66-70.
6. Mehigan JT, Stoney RJ. Arterial microemboli and fibromuscular dysplasia of the external iliac arteries. Surgery 1977;81:484-6.
7. Drury JK, Pollock JC. Fibromuscular dysplasia of the iliac arteries. Vasc Surg 1982;16:133-6.
8. Tsnado J, Barnes RW, Beachly MC, Vines FS, Amendola MA. Fibrodyysplasia of the popliteal arteries. Angiology 1982;33:1-5.
9. Inspall RJ, Chamberlain J, Loose HW. Fibromuscular dysplasia of visceral arteries. Eur J Vasc Surg 1992;6:668-72.
10. Iwai T, Konno S, Hijii K, Satake S, Suzuki S, Hiranuma S, et al. Fibromuscular dysplasia in the extremities. J Cardiovasc Surg (Torino) 1985;26:496-501.
11. Schneider P, LaBerge J, Cunningham C, Ehrenfeld WK. Isolated thigh claudication as a result of fibromuscular dysplasia of the deep femoral artery. J Vasc Surg 1992;15:657-60.
12. Esfahani F, Rooholamini SA, Azadeh B, Daneshbod K. Arterial fibrodyysplasia: a regional cause of peripheral occlusive disease. Angiology 1989;40:108-13.

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