Congenital-idiopathic superficial femoral artery aneurysm in a 7-year-old child

Running Head: Femoral artery aneurysm in a child

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Abstract

Superficial femoral artery aneurysm in children is distinctly uncommon, and usually results from infection, vasculitis, connective tissue disorder, or trauma. We report a 7-year-old girl who had multiple fusiform aneurysms of the right superficial femoral artery, with no evidence of related disorders. The patient successfully underwent aneurysm resection and femoral artery reconstruction with autogenous saphenous vein. Histological examination revealed intimal thickening with fibroplasia without severe inflammatory infiltrates or cystic medial necrosis, suggesting a congenital-idiopathic arterial aneurysm. Three years after the procedure, the saphenous vein graft is fully patent and the patient is in good condition.

Introduction

Femoral artery aneurysms in children are rare. Most cases are associated with infection, vasculitis or trauma, with congenital-idiopathic cases being extremely uncommon. We report the successful treatment of such a case of in a 7-year-old girl.

Case Report

A 20-month-old girl presented with an asymptomatic pulsatile mass in the medial region of her right thigh. There was no familial background of Marfan's syndrome, Ehlers-Danlos syndrome or other autoimmune connective tissue diseases. She had no history of rheumatic fever, Kawasaki disease, trauma or other vascular diseases. A magnetic resonance angiography (MRA) and ultrasound scans showed multiple fusiform aneurysms of the right superficial femoral artery with a total length of 10 cm and a maximal diameter of 3 cm (Fig 1). There was no arterial obstruction, major thrombus or significant development of collateral circulation. The aneurysm showed no signs of compression on adjacent veins or nerves. Considering the child's age, size and lack of symptoms, a definitive repair was postponed to allow her to grow so that

a lager sized repair conduit could be used. However, careful follow-up of every 3 months with attention to the aneurysm size, mural thrombosis, distal embolism and arterial stenosis was required. She had to lead a life escaping the high impact against the aneurysm.

At the age of 7 years (height 120 cm, weight 20 kg) the patient was admitted to our hospital because of the sudden onset of throbbing pain in her right thigh while performing gymnastic exercises.

On physical exam there was no sign of limb ischemia and her peripheral arteries were well palpable. A serological test for inflammation (C-reactive protein and erythrocyte sedimentation rate) was negative.

An MRA demonstrated that the femoral aneurysm was partially filled by a fresh thrombus but had not significantly grown in size when compared to previous studies. Despite this, the patient’s intractable leg pain was worrisome for impending aneurismal rupture and, as a result, the patient was taken urgently to the operating room.

Through a longitudinal incision, the common, deep, superficial femoral arteries and the proximal popliteal artery were exposed and isolated. The common femoral artery and proximal popliteal artery were noted to be

approximately 5 mm and 3.5 mm in external diameter, respectively. A fusiform aneurismal mass 10 cm in length and 3.5 cm in width was found to originate from the most proximal part of the superficial femoral artery. There was no sign of rupture and the aneurysm wall was found to be partially calcified. Upon opening the aneurysm a large amount of fresh thrombus was noted. The aneurysm was then resected and a femoropopliteal bypass was performed using an autogenous reversed saphenous vein graft (3.5-mm in external diameter). The proximal end of the vein graft was anastomosed to the side of the common femoral artery and the distal end to the side of the popliteal artery. Both anastomoses were carried out with a running 7-0 polypropylene suture interrupted at lengths to promote future vascular growth. After completion the graft flow was measured as 90 mL/minute by an electromagnetic flow meter.

Histologic evaluation of a portion of the resected aneurysm revealed irregularly fibrous thickening of intima and attenuation of the elastic fibers of the media by massive fibrosis. Lymphocytic infiltration was focally observed in adventitia; however no feature of vasculitis or infection was found. Cystic medial necrosis was not present. These histological findings were compatible with congenital-idiopathic aneurysm (Fig 2).

Heparin was administered for a week, and warfarin was used for a year postoperatively until fully restoring the endothelial layer, because the vessel was thin. Since discontinuation of anticoagulation, the patient has taken aspirin. A recent MRA and a Duplex ultrasound showed no obstruction or deformation of the graft (Fig 3). Three years after the procedure, the patient continues to be asymptomatic and enjoys unrestricted daily activity.

Discussion

Femoral artery aneurysms are seen mostly in patients over 70 years old.[1] In the pediatric population femoral artery aneurysms are far less common.[2] Congenital-idiopathic peripheral arterial aneurysms in children are extremely rare. In 1991, Sarkar et al[3] reported that only six cases of such aneurysms had ever been described in the literature. Our recent survey of the literature revealed five additional cases of isolated congenital-idiopathic peripheral artery aneurysms (age 3 to 14 years) including two cases in the iliac artery, one in the deep femoral artery, one in the popliteal artery, and one patient with both iliac and femoral artery aneurysms.[4-8] To the best of our knowledge, the present case is the youngest surgically treated patient of superficial femoral artery aneurysm with congenital-idiopathic etiology.

The etiologies of the arterial aneurysms in children are manifold. Sarkar et al[3] reported clinicopathological classification of nine categories for arterial aneurysm in children in detail. These are arterial infection, giant-cell aortoarteritis, autoimmune connective tissue disease, Kawasaki's disease, Ehlers-Danlos syndrome or Marfan's syndrome, other forms of noninflammatory medial degeneration, arterial dysplasias, congenital-idiopathic factors and false aneurysms associated with extravascular events. Considering the history and clinical and pathological signs in our case, the diagnosis corresponded to congenital-idiopathic factors.

The natural history of congenital-idiopathic peripheral artery aneurysm is unknown. As a result, there are no definitive guidelines regarding the timing of repair. Repair in children is further complicated by the technical challenges of small vessels and the need for vascular growth. Therefore, in the absence of symptoms or rapid growth, conservative management is often advocated to allow time for growth.

Choice of graft materials is another concern in children. Use of prosthetic grafts exposes patients to the life-long risk of infection and relative stenosis as they grow. Watelet et al[9] reported in their long-term follow-up adult

femoropopliteal bypass case study that reversed saphenous vein grafts provided better patency than in situ grafts when veins were less than 4mm in diameter. The graft patency for veins larger than 4mm was similar between the two methods. Considering this data, reversed saphenous vein is probably the graft of choice for peripheral vascular reconstruction in pediatric population.

As for the anastomosis, most of them were running anastomosis that were well spatulated and few of them were used the interrupted absorbable monofilament suture technique to allow undisturbed arterial growth in our case because vein graft was large enough to anastomosis. However, if requiring the anastomosis between thin blood vessels, side-to side anastomosis by all interrupted sutures may be better to avoid the purse-string effects of continuous sutures. Saphenous vein bypass also requires careful attention and meticulous preparation. Normothermic graft storage in heparinized whole blood and non-distension of the vein graft during its harvesting and preparation may be key element.

Late aneurysmal degeneration of saphenous vein grafts is known to be another concern in children. Cardneau et al [10] cited that 14.2% of saphenous vein grafts showed late aneurysmal dilatation at an average of 10.7 years after

operation, although the degeneration may be different in both of the aortorenal circulation and the lower extremities. Careful long-term follow-up is necessary with special attention to both stenosis and dilatation when saphenous veins grafts are used in children.

Conclusion

We successfully performed resection and reconstruction with autogenous reversed saphenous vein to treat the impending rupture of a complex fusiform superficial femoral artery aneurysm in a 7-year-old girl.

Figure Legends

Fig 1. Magnetic resonance angiography (MRA) showing a string of fusiform aneurysms. The aneurysm is located in the entire right superficial femoral artery.

Fig 2. Surgical specimen of right superficial femoral artery aneurysm.

a, Irregularly thickened and thin arterial wall with extensive fibrosis (hematoxylin and eosin〔H&E〕stain loupe).

b, Fibrous thickening occurred mainly in the intima (H&E stain x40).

c, Elastic fibers of arterial wall were attenuated or focally disappeared (elastica van Gieson stain x40).

d, Inflammatory infiltrates focally aggregated in the adventitia (H&E stain x200).

Fig 3. Post-operative magnetic resonance angiography (MRA) demonstrating patent graft to a popliteal artery.

References

[1] Corriere MA, Guzman RJ. True and false aneurysms of the femoral artery. Semin Vasc Surg. 2005;18:216-23.

[2] Lund RH. Congenital aneurysm of the common femoral artery. J Pediatr Surg. 1969;4:547-52.

[3] Sarkar R, Coran AG, Cilley RE, Lindenauer SM, Stanley JC. Arterial aneurysms in children: clinicopathologic classification. J Vasc Surg. 1991;13:47-56; discussion 56-7.

[4] Taketani S, Imagawa H, Kadoba K, Sawa Y, Sirakura R, Matsuda H. Idiopathic iliac arterial aneurysms in a child. J Pediatr Surg. 1997;32:1519-21.

[5] Zimmermann A, Kuehnl A, Seidl S, Eckstein HH. Idiopathic aneurysm of the common iliac artery in an 11-year-old child. J Vasc Surg. 2009; 50: 663-6. Epub 2009, July 3.

[6] Rainio P, Biancari F, Leinonen S, Juvonen T. Aneurysm of the profunda femoris artery manifested as acute groin pain in a child. J Pediatr Surg. 2003;38: 1699-700.

[7] Hurley PR, Giddings AE. Idiopathic true aneurysm of the popliteal artery in childhood. Cardiovasc Surg. 1994;2:381-3.

[8] Sheppard DG, Wilkinson AG. Syndrome of idiopathic childhood aneurysms: a case report and review of the literature. J Vasc Interv Radiol. 2000;11: 997-1004.

[9] Watelet J, Soury P, Menard JF, Plissonnier D, Peillon C, Lestrat JP, et al. Femoropopliteal Bypass: in situ or reversed vein grafts? Ten-year results of a randomized prospective study. Ann Vasc Surg. 1997;11:510-9.

[10] Cardneau JD, Henke PK, Upchurch GR Jr, Wakefield TW, Graham LM, Jacobs LA, et al. Efficacy and durability of autogenous saphenous vein conduits

for lower extremity arterial reconstructions in preadolescent children. J Vasc Surg. 2001;34:34-40.

Fig.1

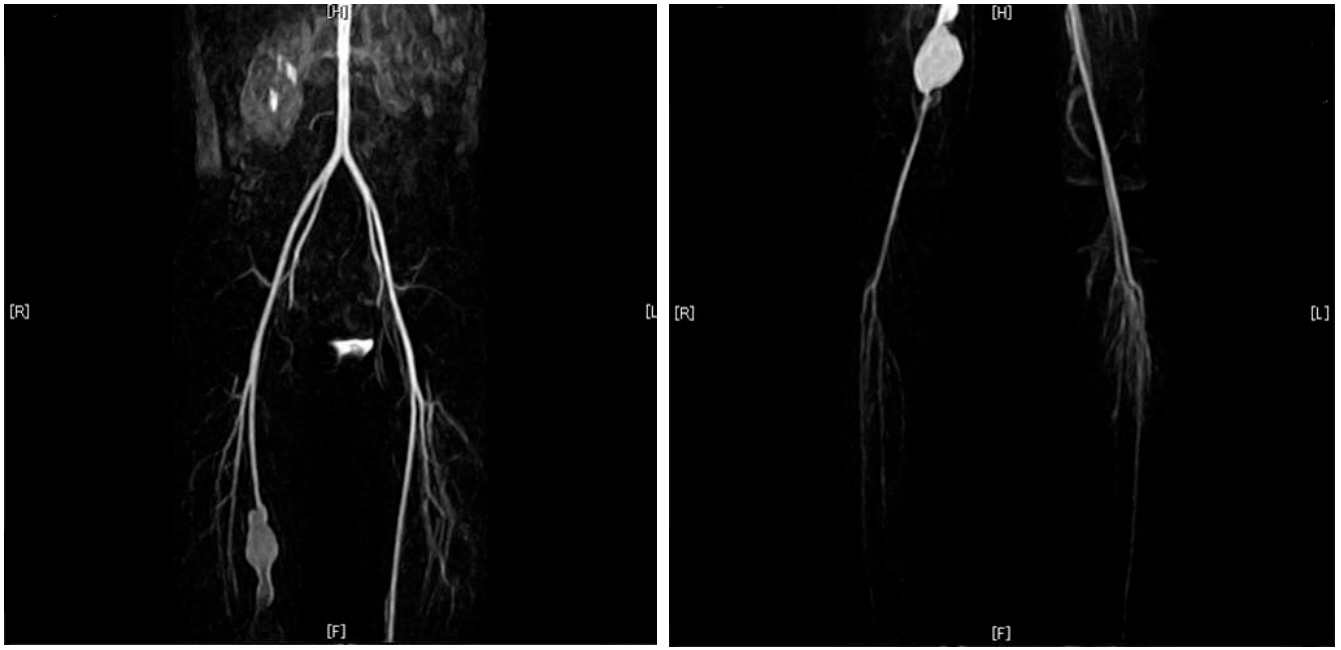
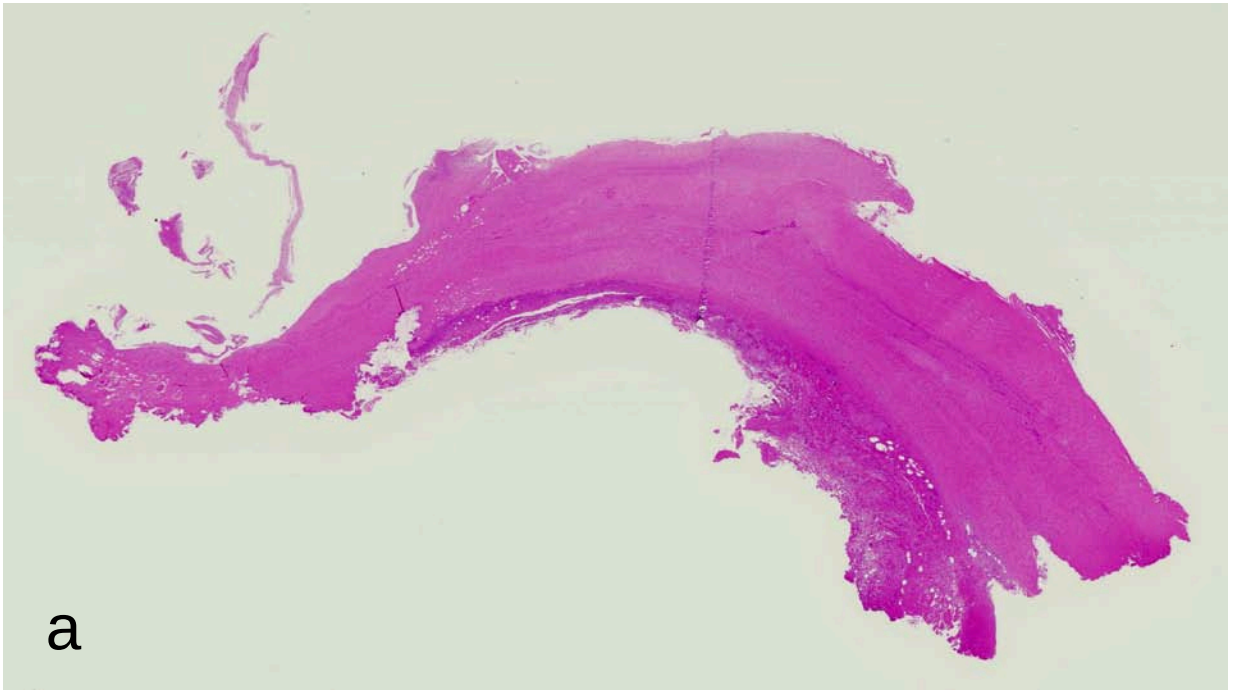
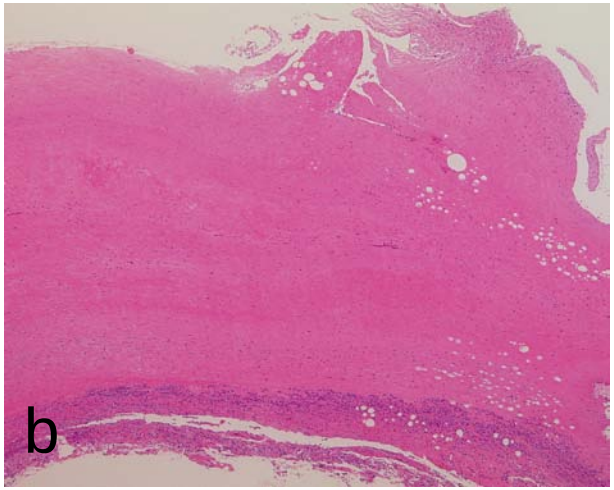
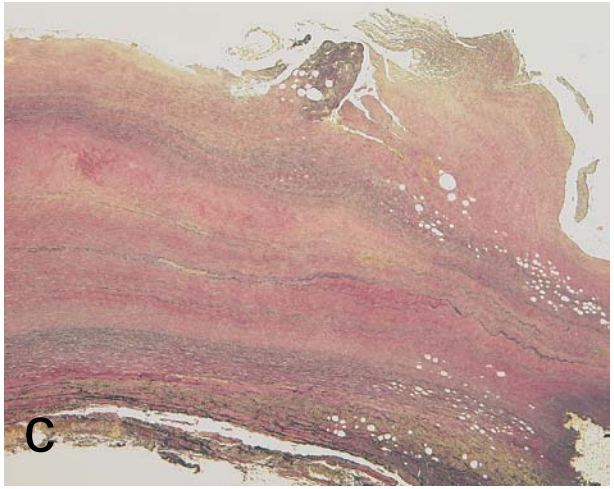


Fig.2







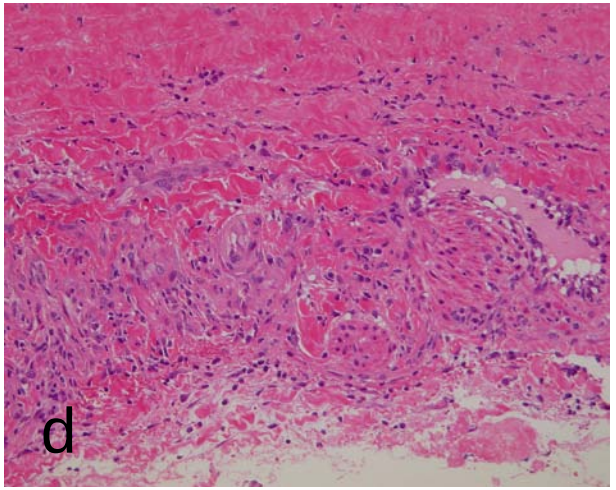


Fig.3