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Popliteal Artery Entrapment Syndrome caused by Spontaneous Rupture of a Popliteal Artery Aneurysm in a 12-year-old Boy

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Most cases of popliteal artery aneurysm occur in the elderly at the age of sixty to seventy years. Atherosclerosis is the most common etiological factor, but other risk factors such as trauma or infection are seen occasionally. Clinically, thrombosis or thromboembolization are often seen, but rupture is particularly rare, especially in children. We report here a robust 12-year-old boy who suffered from a spontaneous popliteal artery aneurysm rupture complicated with critically ischemic leg and compartment syndrome. We performed emergency surgery for reconstruction of the popliteal artery with the autologous saphenous vein followed by fasciotomy. After the operation, limb salvage was achieved and the patient now has almost normal lower limb activity one year later.

Key words: popliteal artery aneurysm, rupture, autologous saphenous vein, fasciotomy

INTRODUCTION

Popliteal artery aneurysm is infrequent but is the most common of all peripheral aneurysms[1,2,7]. It usually occurs in the elderly at the age of sixty to seventy years, whereas it seldom occurs in children. It is often misdiagnosed, because it is usually clinically asymptomatic and the physical examination of the popliteal region is difficult[3,6]. A popliteal artery aneurysm should be considered if a patient suffers from lower leg pain without definitive causes[8]. Thrombosis and distal embolization producing severe ischemia of the lower extremity are the most frequent and well-known complications of this condition[2]. An acute rupture of popliteal artery aneurysm is uncommon but is particularly dangerous because of the threat of loss of the lower leg and occasionally of death[3]. Here we report a 12-year-old boy who suffered from a spontaneous rupture of a popliteal artery aneurysm and detail its treatment.

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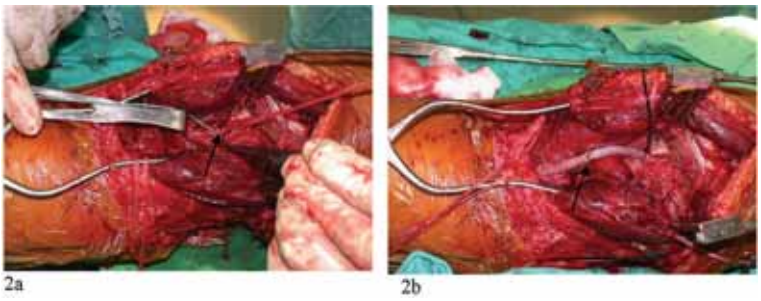
CASE REPORT

A 12-year-old boy was brought to our emergency department by his parents in September 2006 with a seven-day history of progressive right lower leg swelling. According to the statements of the patient and his parents, he started to suffer from right lower leg swelling at school. However, there was no history of trauma. At first, they did not pay much attention to it until progressive pain and a swollen leg, even claudication, was noted. Because the symptoms persisted and got worse, he was brought to our emergency department for further evaluation.

A physical examination showed that the patient was a normally developed 12-year-old boy in moderate distress. There was no evidence of congenital diseases or other underlying medical ailment and the family history was noncontributory. Examination of the right lower leg demonstrated tenderness and swelling of the calf and the absence of popliteal and pedal pulses. Laboratory data showed leukocytosis (white blood count 13,000/ μL), anemia (Hgb 9.9 g/dL), elevated CRP (6.77 mg/dL) and elevated CK (16,247 U/L). A plain X-ray film of the lower leg showed normal bone structure and swollen soft tissue around the popliteal fossa. Computerized tomographic angiography revealed severance of the popliteal artery below the knee (Fig. 1). Emergency surgery using reconstruction of the artery with the autologous saphenous vein was performed (Fig. 2). Following reconstruction of the popliteal artery, fasciotomy was applied to treat the compartment syndrome. Histology of the specimen showed disruption of the entire vascular wall, thinning of the tunica

Fig. 1 Computerized tomogram with graphic reconstruction demonstrates the severance of the popliteal artery below the knee (black arrow).



2a 2b

Fig. 2 (a) The ruptured popliteal artery aneurysm was identified (black arrow) , and (b) reconstruction of the distal perfusion with the autologous sapheneous vein was performed (black arrow).

media and fibrin thromboses with hemorrhage. Rupture of a popliteal artery aneurysm was thus confirmed.

After the surgery, massive fluid supplementation was given to maintain a satisfactory urine output (>100 mL/h). Intravenous anticoagulation (heparin) was used. Prostaglandin E1 was also added to increase peripheral blood flow. Patency of the arteries in the right lower leg was verified using a bedside Doppler ultrasound scan. The patient's distal leg perfusion improved gradually and pulsation of the dorsal pedis artery became palpable. After adequate wound debridement and split-thickness skin graft coverage over the fasciotomy region, limb salvage was achieved. After an adequate rehabilitation program, the boy has almost normal lower limb activities one year later.

DISCUSSION

Popliteal artery entrapment syndrome (PAES) is a rare cause of lower extremity arterial insufficiency in young adults. Most frequently, the syndrome is seen in a well-developed man in his twenties or thirties with intermittent claudication of the calf muscles[10]. In pediatric patients, ischemic symptoms can be misinterpreted as musculoskeletal pain[11]. Chronic trauma and aberrant flow dynamics can lead to aneurysmal change or thrombosis of the popliteal artery, resulting in limb-threatening ischemia[10]. Early and correct diagnosis is important because PAES is a progressive condition and early treatment of the symptomatic patient may prevent devastating vascular complications or limb loss[11]. PAES rarely happens in children and popliteal artery aneurysms in children tend to be found incidentally. Popliteal aneurysms are infrequent but nevertheless the most common of all peripheral aneurysms[3]. They occur primarily in elderly people and most of the patients are aged 60 years or older[1]. They are clinically very significant, as they can lead to limb threatening situations and are recognized as the "silent killer of the leg circulation"[7]. About half of all popliteal aneurysms are bilateral and accompanied by aortic or femoral aneurysms[9]. Patients might be asymptomatic or can present acutely with thrombosis or peripheral embolization[9]. Ruptured popliteal artery aneurysms are rare, especially in children[2,9]. A rupture can present as a massively swollen leg with loss of blood and should be suspected if no other causes are evident[4]. If one fails to recognize this limb-threatening condition, it could lead to delay in treatment and even limb loss[4]. Besides, because of the strong musculofascial compartment surrounding the popliteal artery, the expanding hematoma can obstruct the corresponding veins[8]. Compartment syndrome can be found following a ruptured popliteal artery aneurysm. Therefore, early recognition and immediate treatment are critically important.

The timely diagnosis of a ruptured popliteal aneurysm is undoubtedly the most difficult aspect in management. Clinically, they should be suspected in elderly patients presenting with symptoms or signs of generalized atherosclerosis and in the popliteal fossa. Noninvasive techniques including color duplex scanning, computerized tomography and magnetic resonance imaging have been advocated to evaluate the popliteal space and to confirm the diagnosis of aneurysmal rupture. However, if time permits, angiography is necessary to provide information

regarding the vessels proximal and distal to the aneurysm so that appropriate surgery can be planned. Early surgical repair is the mainstay in the management of ruptured popliteal aneurysms.

Two goals of effective surgery are the control of bleeding and reestablishment of distal perfusion[2]. Five-year graft patency rates after surgical repair have ranged from 30% to 97%, with five-year limb salvage rates ranging from 70% to 80%[7]. In nonruptured popliteal aneurysms, endovascular management has replaced open surgical repair in some situations. However, the durability of stents in such cases needs further follow-up to evaluate the long-term results[7,9]. An acute rupture of an aneurysm of the popliteal artery is relatively rare and represents less than 4% of all such cases[3]. Here we described a young boy with a spontaneous rupture of a popliteal artery aneurysm who had critical limb ischemia because he and his carers were unaware of the severity of the condition. Increased awareness of this condition and its clinical presentation is of utmost importance, despite its rarity.

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