

Sequential development of multiple atherosclerotic peripheral arterial aneurysms in a four-year period

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Abstract

Compared to the other isolated peripheral aneurysms, multiple peripheral aneurysms are less commonly encountered. We present a rare case of multiple aneurysms involving subclavian, bilateral popliteal and iliofemoral arteries which developed sequentially over a 4-year period. Histopathological studies of the specimens obtained from aneurysmal sacs demonstrated a correlation with atherosclerotic aneurysm. The aneurysms of the patient were successfully operated on sequentially at different times by using various surgical procedures. This case demonstrated that these patients should be monitored closely during their lifetime for development of new arterial aneurysms at different sites.

Key words: aneurysm, atherosclerosis, peripheral vascular disease.

Introduction

The peripheral aneurysms are considered as potential limb threatening conditions, but appropriate diagnosis and treatment can avoid possible complications. The development of more than one arterial aneurysm in the same individual is relatively uncommon but all the same it may occur if the same disease processes which had caused the initial lesion continue to exist. In the present case report, clinical presentation of multiple peripheral aneurysms and the details pertaining to surgical reconstruction of this pathology are described.

Case report

A 45-year-old hypertensive man exhibiting numbness in the left arm, pain associated with exercise, and a left supraclavicular mass which had been continuously increasing in size for the past 3 months was admitted to our clinic initially in July 2000. The patient had no history of trauma, drug abuse, or infective endocarditis and familial history of aneurysms; he had had essential hypertension for 3 years and had been smoking a pack of cigarettes each day for the past 25 years.

Physical examination revealed a pulsatile mass in the left supraclavicular fossa. Left radial, ulnar and brachial artery pulses were weak. Neurological examination showed a mild loss of sensation in the left arm. Examination revealed no abnormality in other systems. There were no signs of connective tissue diseases associated with arterial involvement such as hyperelastic skin, hypermobile joints, or marfanoid habitus. Laboratory examinations including erythrocyte sedimentation rate, C reactive protein, complete blood



Figure 1. Digital subtraction angiography image of left subclavian artery aneurysm

count, serological test for syphilis, rheumatoid factor, antinuclear antibody, antithrombin III, protein C, and protein S, were normal except hypercholesterolaemia. A colour Doppler ultrasound suggested the presence of a subclavian aneurysm. Combined peripheral and coronary arterial angiographic examination revealed a 5 × 7 cm aneurysm at the middle portion of the left subclavian artery (Figure 1) along with normal coronary and other peripheral arterial angiographic findings.

The patient underwent an operation and a clavicular resection was performed through a supraclavicular incision. The aneurysm was explored and excised after ligation of the proximal end of the subclavian artery. Despite clavicular resection, a left carotid to subclavian extra-anatomic bypass was performed using an autologous saphenous vein graft due to anatomical and surgical difficulties. The postoperative course was uneventful. The patient was discharged on the 8th postoperative day. The patient was followed up periodically by our clinic after the first surgical operation. He was readmitted to our clinic in June 2003 due to pain in the right leg and swelling in the right groin. In the physical examination, pulsatile masses were identified in the right groin (3 × 5 cm) and in the right and left popliteal areas, 4 × 6 cm and 3 × 3 cm, respectively; and right distal leg pulses were non-palpable. Aneurysmal enlargements in the regions of the anatomical locations mentioned above were demonstrated with magnetic resonance angiography (MRA) (Figure 2). Our patient underwent a second operation; the right femoral aneurysm was excised and iliofemoral graft interposition was performed with an 8 mm polytetrafluoroethylene graft. The right popliteal artery aneurysm was excluded from the arterial circulation by proximal and distal ligatures. Following a selective distal embolectomy, interposition of a femoro-popliteal graft with the saphenous vein was performed. Approxi-

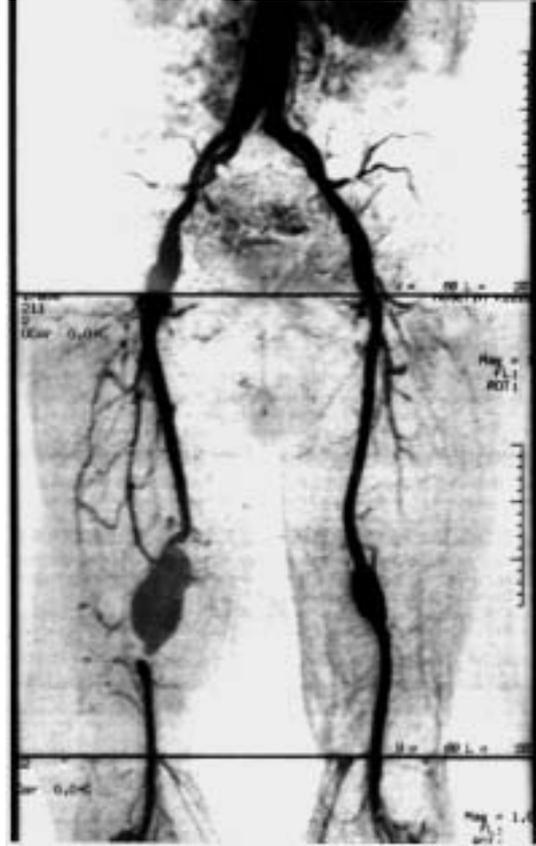


Figure 2. Magnetic resonance angiography of the lower extremities with arterial aneurysms

mately 3 months after the second operation, he underwent a third operation for a left popliteal mass. The aneurysm in the left popliteal region was excised and a saphenous vein graft interposition was performed (Figure 3). Thrombosis of the iliofemoral graft was identified in the right leg 6 months after the second operation, and graft thrombectomy was performed.

In this patient, all aneurysms were observed as true aneurysms. Histopathological studies of the specimens obtained from aneurysmal sacs demonstrated a correlation with atherosclerotic aneurysm. The walls of the resected aneurysms were ulcerated; moreover, thrombosis and hyaline degeneration along with fibrosis of the media layer with lymphohistiocytic infiltration were also determined (Figure 4). These histological findings indicated atherosclerosis as the underlying cause of the aneurysms.

After the surgical procedures, his dietary intake was restricted with a new dietary programme and he was subjected to antihyperlipidaemic (atorvastatin), antiplatelet (aspirin) and antihypertensive (carvedilol) therapies. The patient has been periodically followed up with arterial Doppler ultrasonography and MR angiography for about

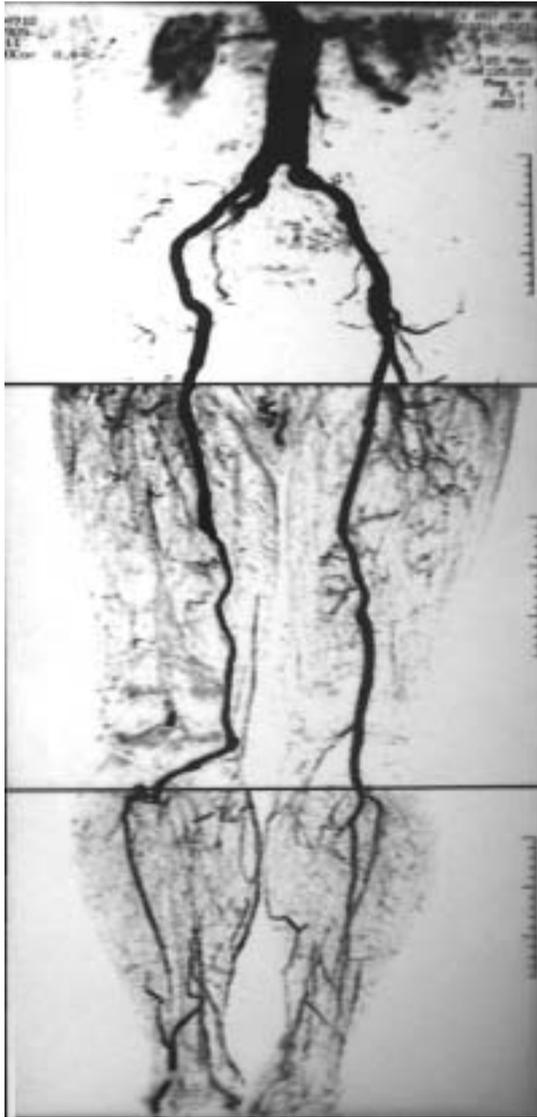


Figure 3. Control MR angiography image of the lower extremity arteries 12 months after the last operation

3 years. The vascular status of the lower and upper extremities were still normal despite the weakening distal arm pulses of the upper extremities in the course of time. But those were not serious enough to cause ischaemic symptoms. We have not determined any new aneurysmal development.

Discussion

Peripheral arterial aneurysms (PAAs) are relatively uncommon compared to the abdominal aortic aneurysms. But the current incidence of non-specific peripheral aneurysms appears to be on the rise due to an increase in age range of the general population and advances in imaging techniques [1]. In a series reported in 1972, the ratio of peripheral atherosclerotic aneurysms to abdominal aortic aneurysms was shown to be 1 : 23 [2]. More recent studies revealed the ratios

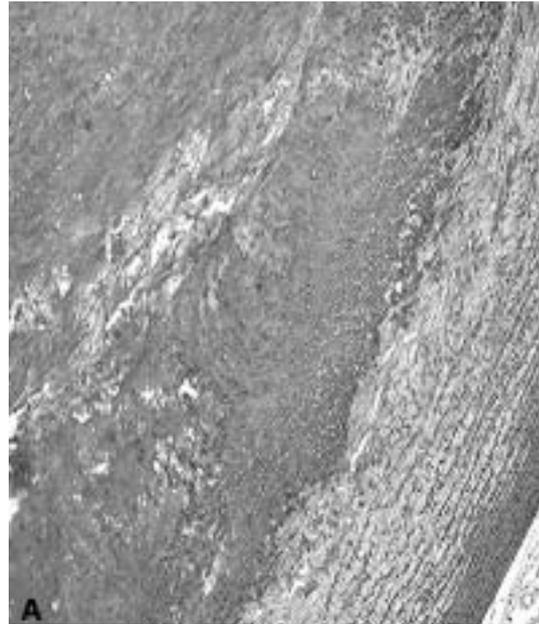


Figure 4. Histological sections of the walls of the aneurysms, exhibiting typical atherosclerotic changes. (A) Intimal ulceration and thrombosis, H&E, 40× magnification. (B) Degeneration and fibrosis of media layer with lymphohistiocytic infiltration, H&E, 200× magnification

of popliteal aneurysms and abdominal aortic aneurysms as 1 : 15 and 1 : 8 [3, 4]. Patients with subclavian arterial aneurysm should be thoroughly investigated for presence of other associated aneurysms, especially in cases of atherosclerosis. In a review of the literature related to subclavian artery aneurysm, Dougherty *et al.* demonstrated that 33-47% of patients with subclavian aneurysms had concomitant aortic, visceral and peripheral

aneurysms [5]. On the other hand, in patients diagnosed with popliteal aneurysms, the probability of encountering multiple aneurysms is known to be very high. Popliteal arterial aneurysms are bilateral in 40 to 60% of cases [6]. The great majority of these patients are expected to have an extrapopliteal aneurysm, as well. The most common sites are in the femoral (38-48%) and aortic (36-58%) positions, and they may occur in up to 78% of cases in the presence of bilateral popliteal involvement [7]. Multiple aneurysms are clinically common among the elderly population aged above sixty, and are caused mainly by atherosclerosis [8]. In young patients, other aetiologies that could be responsible for the development of arterial aneurysms should be considered, as well [8]. Connective tissue disorders such as Ehlers-Danlos syndrome, neurofibromatosis type-1, and Marfan's syndrome and arteritis such as Behcet's syndrome, Kawasaki disease, and giant cell arteritis have also been associated with peripheral aneurysms, mostly in children or young adults [9]. In idiopathic aneurysms, the role of matrix metalloproteinases and their tissue inhibitors in the aetiology of vascular connective tissue destruction is increasingly recognized [10]. However, the present case may suggest an underlying defect in collagen metabolism; he had no characteristic findings for genetic disorders such as Ehlers-Danlos or Marfan's syndrome which could be detected with genetic testing. Histopathological studies of the specimens obtained from aneurysmal sacs demonstrated a correlation with atherosclerotic aneurysm.

Regardless of the aetiology, the principles of surgical treatment for the aneurysms are the same. Emergency treatment is indicated in the presence of limb-threatening complications such as embolization, thrombosis and rupture [1, 11]. Until recently, surgery was the golden standard of treatment for all aneurysms. However, lately, endovascular repair of a PAA has also been proven to be a viable treatment option [12, 13]. Surgical operation is indicated in all of the symptomatic and complicated aneurysms. Patients with local findings such as continuous increase in pain, and venous or neural complications, should be treated without delay. Although the surgical criteria for asymptomatic arterial aneurysms are not very clear, many surgeons also recommend surgical operation in all asymptomatic patients, except for high-risk cases [1, 14]. The surgical principles are the same regardless of the location of the aneurysm: the aneurysms should be excluded and arterial circulation must be restored.

In conclusion, we suggest that in patients with peripheral aneurysms, other possible aneurysms should be investigated by applying appropriate techniques, and that these patients should be

monitored closely during their lifetime, for they continue to live under the risk of developing complications related to new aneurysms in other sites.

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