Vasospastic limb ischemia might have been underappreciated compared to vasospasm in other territories such as heart and brain. However, an increasing awareness of this vascular disorder can be translated to an improved patients’ care. Herein, we report a case of vasospasm presenting acute and chronic limb ischemia in four extremities.

Keywords: vasospasm, limb ischemia, recurrence

Introduction

Vasospasm in the extremities is uncommon compared to vasospasm in other territories such as heart and brain. However, an increasing knowledge about vasospastic limb ischemia is crucial in appropriate diagnosis and treatment for vascular specialists. Herein, we report a case of vasospasm presenting both acute and chronic limb ischemia in four extremities.

Case Report

A 28-year-old male (height 182 cm, body weight 80 kg) with a current smoking history was referred to our hospital for the diagnosis and treatment of developing ischemic symptoms in upper and lower limbs. Two weeks before, the patient had experienced bilateral hand pain at rest. One week before, the patient’s symptom had included life-style limiting claudication and calf pain at rest despite smoking cessation. On his arrival, peripheral pulsations in radial arteries, dorsalis pedis arteries, and posterior tibial arteries were absent. The patient’s blood pressure was 121/60 mmHg and pulse rate was 82 beats per minute with regular rhythm. Laboratory data including lipid and diabetic profile was normal except for increase in serum creatinine phosphokinase (3961 U/L). Chest X-ray and electrocardiography also revealed no abnormality. Diagnostic imaging including duplex ultrasonography and enhanced computed tomography (CT) demonstrated the artery disruptions in upper and lower extremities (Fig. 1A). We initiated medical treatment including intravenous administration of alprostadil as well as oral administration of cilostazol and beraprost immediately after hospitalization. His symptom dramatically improved with significant recovery of pulsation in extremity arteries, and imaging examinations also demonstrated excellent recovery of disruptive lesions (Fig. 1B). The patient uneventfully discharged under medical management 7 days later. A rheumatologic and serological examination proved negative, and neither relationship between the patient’s symptom and the cold stimulus suggestive of Raynaud’s disease nor occupational history inducing vibration syndrome and hammer syndrome
Kaneyama J, et al. was rehospitalized for the treatment of “acute” limb ischemia due to recurrent vasospasm. Oral administration of calcium channel blocker and isosorbide dinitrate-containing patch were added to the baseline treatment. His symptoms dramatically improved with full recovery of four limb arteries’ pulsation immediately after medical treatment. Enhanced CT also detected successful recovery of the disruptive arteries (Fig. 2B). Although serum creatinine phosphokinase increased up to 40820 U/L and reperfusion injury following the relief of vasospasm was observed in bilateral calves (Fig. 3), neither of myonephropathic metabolic syndrome, compartment syndrome nor amputation was observed. He was uneventfully discharged 16 days later, and no recurrence of vascular event has been observed under medical treatment and smoking cessation during the 12-month follow-up period.

Discussion

To the best of our knowledge, this is the first to report recurrent limb ischemia consisting of not only chronic limb ischemia but also acute limb ischemia due to spontaneous vasospasm in four extremities.

Vasospasm can occur by the constriction of arterial smooth muscle in the territory of muscular artery. Drug-induced vasospastic limb ischemia by ergotamine, methysergide, cocaine and lysergic acid diethylamide is well known.4–6) However, there are only a few cases regarding spontaneous vasospasm in the lower limb artery in which acute onset of femoropopliteal or peroneal artery occlusion was observed.1,2) Unfortunately, the reason for this rare phenomenon remains unclear. Of great interest, our case presented both “acute” and “chronic” limb ischemia. The diagnosis of vasospastic limb ischemia could be challenging in the setting of clinical practice. The process of differential diagnosis ruled out other vascular diseases based on the following reasons: (1) lack of atherosclerotic risk factors except for current smoking history; (2) no evidence of vasculitis; (3) no suggestion of Raynaud’s disease; (4) no occupational exposure; (5) no history of drug use to induce vasospasm; and (6) failure to meet diagnostic criteria for Buerger’s disease. Thus, we believed that this patient had vasospastic limb ischemia.

As of today, no established remedy exists despite vasospastic limb ischemia could potentially results...
Vasospastic Limb Ischemia

Thus, the combination of smoking cessation and medical therapy could be the main-stream in the management of vasospasm although further investigation needs to be undertaken regarding optimal medical treatment for vasospastic limb ischemia.

With the possibility of vasospasm in our mind, the appreciation of clinical and angiographical findings can definitely reduce the chance of misdiagnosis and unnecessary invasive intervention, and can be catastrophic clinical scenarios. Smoking is well known to be associated with coronary vasospasm.7) In this case as well, smoking might have served as a trigger of limb arteries’ spasm and smoking cessation could be of help for prevention of recurrence of limb arteries’ spasm. Also, calcium channel blocker can be essential in the medical treatment of coronary vasospasm.8,9) In this case as well, besides vascular dilators, calcium channel blocker might have worked well in terms of prevention of recurrent vasospastic limb ischemia. Thus, the combination of smoking cessation and medical therapy could be the main-stream in the management of vasospasm although further investigation needs to be undertaken regarding optimal medical treatment for vasospastic limb ischemia.

Fig. 2 Enhanced computed tomography (CT) angiogram in the bilateral lower extremity. (A) Diagnostic enhanced CT angiogram showing tight narrowings in the bilateral femoropopliteal segments (large arrows) and disruptions in the bilateral anterior tibial arteries (small arrows). Also, the proximal segment in the superficial femoral artery seems to be spastic. (B) After the initiation of medical treatment in the 2nd hospitalization, complete recovery of those disruptive lesions was observed though crural arteries were superimposed upon the veins.

Fig. 3 Reperfusion injury following the relief of vasospasm in the lower limb arteries. Please note swollen and reddish calves (arrows). (A) right, (B) left.
translated to an improved patient’s care. We emphasize the need for a more awareness of vasospastic limb ischemia in the field of vascular medicine.

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Disclosure Statement
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