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Case report

A rare cause of acute limb ischemia of both upper and lower limbs caused by prolonged vasospasm

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ABSTRACT

Acute limb ischemia is a life-threatening condition caused by various etiologies including atherothrombosis and peripheral embolization. However, in young adults other etiologies should also be considered. We report a rare case of a 19-year-old man developing acute limb ischemia of both upper and lower limbs due to prolonged vasospasm.

Introduction

Acute limb ischemia (ALI) represents a severe and life-threatening condition caused by various etiologies including atherothrombosis and peripheral embolization. Thrombosis in situ develops in regions of pre-existing stenosis in native arteries or in association with previous endovascular or surgical treatment. In the case of embolization, the thrombi originate most often from the left cardiac chambers or large atheromas of the aorta. Less frequent causes include embolic masses arising from arterial aneurysms (e.g. aneurysm of the abdominal aorta or popliteal artery), paradoxical embolism via a patent foramen ovale or atrial septal defect, or direct vascular involvement such as in Buerger's disease, vasculitis, vascular trauma, dissection, entrapment syndrome and others. Association of acute limb ischemia and prolonged vasospasm is considered to be very rare.

Case report

We report the case of a 19-year-old man with a history of tobacco use, cannabinoid abuse, and a previously documented

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episode of methamphetamine intoxication, who was referred to our center for acute ischemia of both upper and lower extremities. The patient's medical history was unremarkable. Except for smoking, he did not have any other traditional risk factors for atherothrombosis. Four days prior to admission, the patient experienced pain and coldness in his left upper limb. He attributed the sensation to inappropriate sleeping position. Several hours later, he noticed the same symptoms in his right upper limb and both lower limbs. The patient was then examined in another hospital where CT angiography was performed and showed conical narrowing of the brachial arteries with distal occlusion (Fig. 1). The patient was then referred to our center for further assessment.

On admission, it was not possible to measure the patient's blood pressure and he was pulseless at the brachial and femoral arteries. A hand-held Doppler failed to detect any signal on any of the peripheral arteries of the upper and lower extremities. All four of his limbs were pale and cold with decreased motor function and sensory perception. The finding was consistent with SVS (Society for Vascular Surgery) class IIb acute limb ischemia. Laboratory evaluation of blood samples and urine was unremarkable except for a mild increase in creatine kinase (398 U/l), myoglobin (116 μg/l), and D-dimers (320 μg/l).

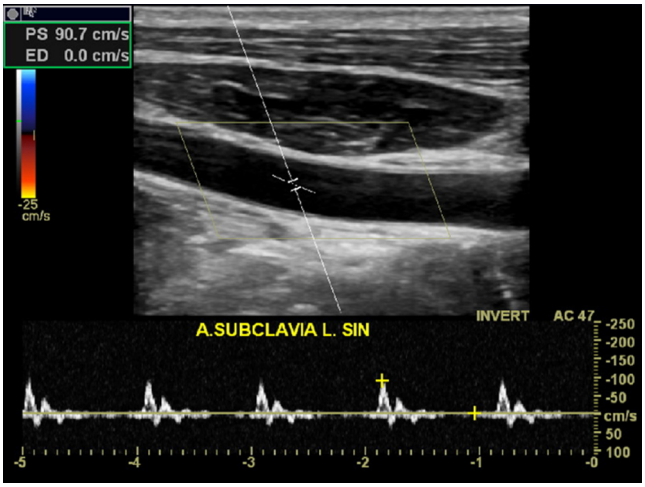

Fig. 2 – PW – Doppler of the left subclavian artery. Duplex ultrasonography with PW – Doppler showing an increased pulsatility in the left subclavian artery.

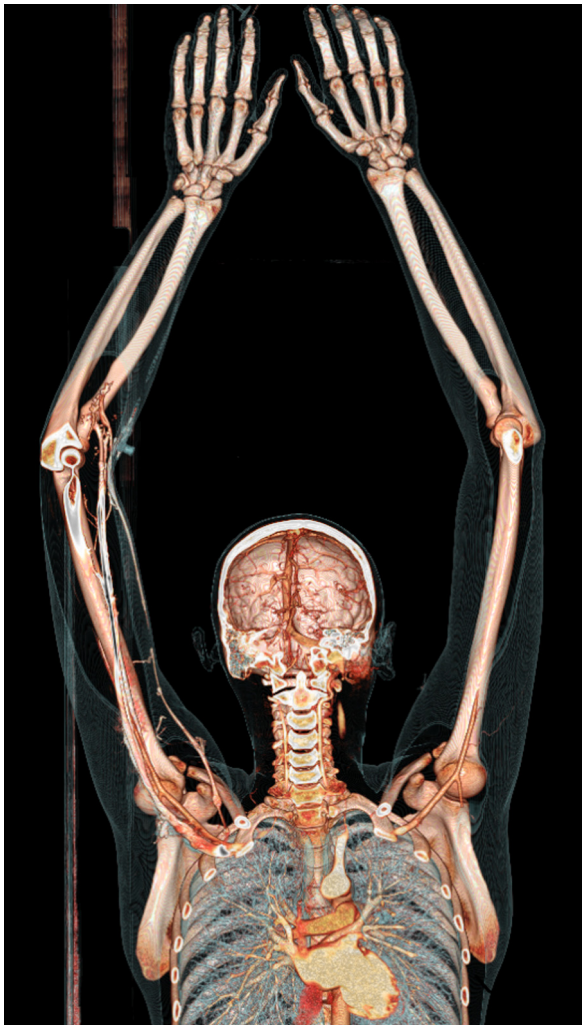

Fig. 1 – CT angiography of the brachial arteries. CT angiography showing conical narrowing of the brachial arteries with distal occlusion, without signs of arterial trauma or dissection.

Considering that perfusion of all four extremities was impaired, multiple embolization was highly unlikely. Duplex ultrasonography with pulse-wave Doppler showed an increased pulsatility in the axillary and femoral arteries with narrowing in their distal parts without signs of atherosclerotic involvement or thrombosis on 2D ultrasound images (Figs. 2 and 3). Arterial angiography was not performed because of patient preference and progressive improvement of symptoms with conservative treatment.

Parenteral anticoagulation therapy with unfractionated heparin was initiated to prevent worsening of ischemia in accordance with current guidelines on the treatment of acute limb ischemia [1].

Transesophageal echocardiography ruled out the presence of intracardiac thrombosis and interatrial shunt. The thoracic aorta appeared normal. Laboratory blood tests focused on the possibility of a hereditary coagulation disorder including factor V Leiden and prothrombin mutations were negative. Levels of protein C and S were also normal. The absence of intracardiac thrombosis and the presence of sinus rhythm throughout hospitalization made cardioembolic etiology highly unlikely.

A panel of the most common autoantibodies (namely rheumatoid factor, antiphospholipid antibodies, ANCA, ANA, ENA, C3, and C4 levels) was performed to rule out autoimmune disease, and showed negative results. Based on these results and normal values of C-reactive protein (2 mg/l) and erythrocyte sedimentation rate (4 mm/h), a diagnosis of vasculitis was highly unlikely. A toxicology screen and urinalysis did not confirm use of illicit drugs. However, urine samples were not obtained until several days after the onset of symptoms. The patient had not been exposed to cold or vibrations and denied symptoms suggestive of Raynaud's phenomenon. The patient

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