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청색 발가락 증후군: 증례 보고

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Blue Toe Syndrome: A Case Report

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Purpose: Blue toe syndrome consists of blue or purplish toes in the absence of a history of obvious trauma, serious cold exposure, or disorders producing generalized cyanosis. It is a life-threatening and still underrecognized disease. It can be commonly occurred by vascular surgery, invasive cutaneous procedures or anticoagulant therapy. Our case is presented of blue toe syndrome related to atheromatous embolization that was presumably triggered by angio CT.

Methods: A 69-year-old man presented with the suddenly developed pain, cyanosis and livedo reticularis of the toes in right foot. Dorsalis pedis pulses were palpable. He had been performed a diagnostic angio CT 1 month earlier. Angio CT revealed diffuse aortic atheromatous plaque in lower abdominal aorta and both common iliac artery. One month after angio CT, he visited our clinic. There was no visible distal first dorsal metatarsal artery and digital artery of right first toe in lower extremity arteriography. A diagnosis was established of blue toe syndrome. Because his symptom was aggravated, we performed the exploration of the right foot. After exposure of first dorsal metatarsal artery, microsurgical atheroembolectomy was done.

Results: There were no postoperative complications. After three months the patient had no clinically demonstrable problems.

Conclusion: Patient with blue toe syndrome is at high risk of limb loss and mortality despite treatment. Blue toe syndrome produces painful, cyanosed toes with preserved pedal pulses. It needs to be aware of blue toe syndrome. Careful history should reveal the diagnosis. Treatment is controversial, however, most believe that anticoagulation therapy should be avoided.

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Key Words: Atheroemboli, Blue toe syndrome

I. INTRODUCTION

Blue toe syndrome was first described in a publication that defined the entity as the sudden onset of acute pain and cyanosis in one or more toes.[1] It may be defined as the development of blue or violescent discoloration of one or more toes in the absence of obvious trauma, serious cold-induced injury, or disorders producing generalized cyanosis.[2] The pathognomonic triad of blue toe syndrome are foot pain, livedo reticularis, and intact pedal pulses.[2]

Atheroemboli producing blue toe syndrome originate from an ulcerated plaque located in the aorta, iliac and femoropopliteal arterial system.[3] This atheroembolism process can happen anywhere in the body. When it occurs in the lower extremities, end arteriole is occluded, and blue toe syndrome is manifested. Once ischemia becomes pronounced, ulceration, gangrene and necrosis can develop and sometimes necessitate limb amputation.

Spontaneously occurred blue toe syndrome was rare.[4] Instead, it is more commonly happened as an iatrogenic problems, especially those caused by injury to the arterial wall from vascular surgery or invasive percutaneous procedures, such as diagnostic angiography and angioplasty.[4,5] Also, various risk factors, such as heparinization, thrombolytic therapy, surgical operations, can cause blue toe syndrome.[3,6]

Blue toe syndrome has been regarded as a problem with a wide spectrum of clinical features, ranging from cyanotic toe to death.[7] Due to high morbidity and mortality, blue toe syndrome should demand prompt recognition and treatment.

We describe a case of the blue toe syndrome presumably due to atheroembolization of atheromatous material from the lower abdominal aorta or common iliac artery is triggered by angio CT.

II. CASE

A 69-year-old man presented with the suddenly developed intense pain in right foot. He was a non-smoker. Examination showed cyanosis and livedo reticularis of the toes (Fig. 1). Dorsalis pedis pulses were palpable and easily audible with a portable doppler. In his past medical history, we were informed that a diagnostic angio CT had been performed 1 month earlier because of skin defect in right 5th toe. Angio CT revealed diffuse aortic atheromatous plaque, multiple luminal irregularity and vascular calcifications in lower abdominal aorta and both common iliac artery. There was no evidence of arterial stenosis in the both lower extremity (Fig. 2). There was no distal arterial embolization after the procedure. The skin defect of 5th toe was healed by conservative treatment.

Laboratory examination revealed an increased level of c-reactive protein (0.30 mg/dl) and erythrocyte sedimentation rates (41 mm/hr), but other findings were normal. Echocardiogram was normal. Both lower extremity arteriography was done and there was no visible distal first dorsal metatarsal artery and digital artery of right first toe (Fig. 3). A diagnosis was established of blue toe syndrome. Because his symptom was virtually aggravated, we performed the exploration of the right foot. The first dorsal metatarsal artery was exposed through a zig-zag dorsal skin incision. Double approximator clamp was placed at the proximal portion and distal portion of the embolus lodged at the first dorsal metatarsal artery. An arteriotomy of the involved first dorsal metatarsal artery was then performed over the embolus. From this arteriotomy, the embolus was extracted using microforceps. Then the proximal clamp was removed and a proximal portion of the first dorsal metatarsal artery showed a good bleeding via an arteriotomy. After applying the proximal clamp, the arteriotomy was then

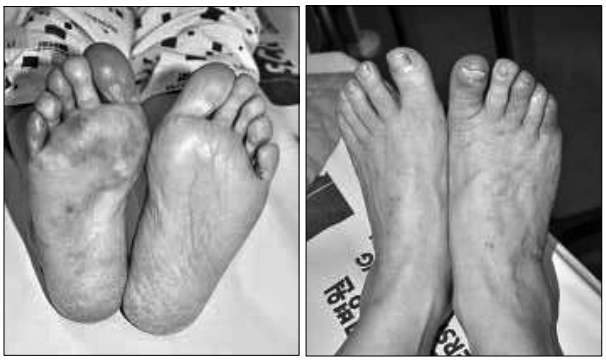

Fig. 1. Violescent discoloration of the toes in a patient with blue toe syndrome.

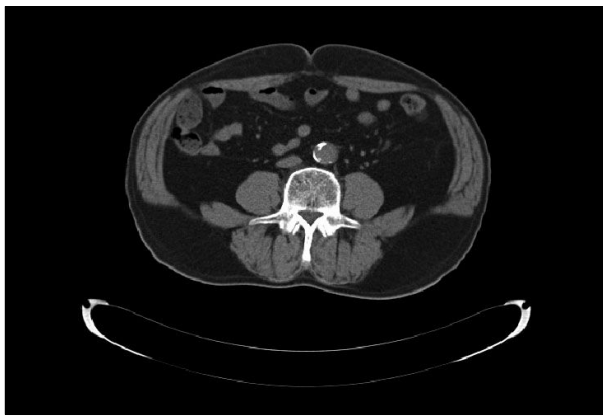

Fig. 2. Angio CT revealed a characteristic findings in blue toe syndrome that embolized was irregular thrombus overlying ulcerative atheromatous plaques.

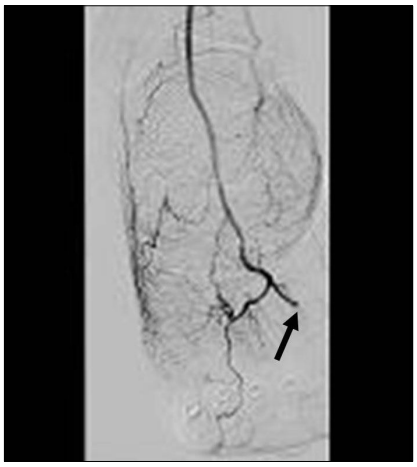

Fig. 3. Lower extremity arteriography was showed that there was no visible distal first dorsal metatarsal artery and digital artery of right first toe (arrow).

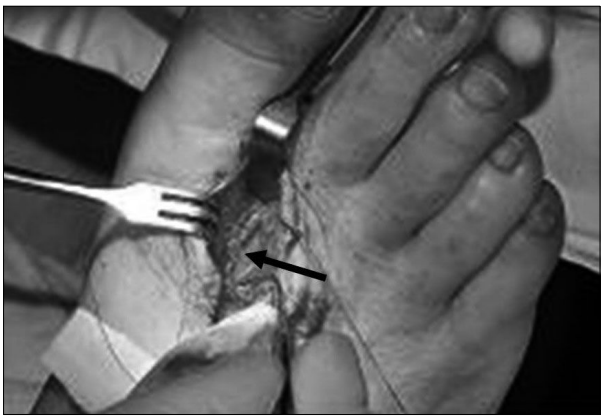

Fig. 4. After exposure of first dorsal metatarsal artery (arrow), microsurgical atheroembolectomy was done.

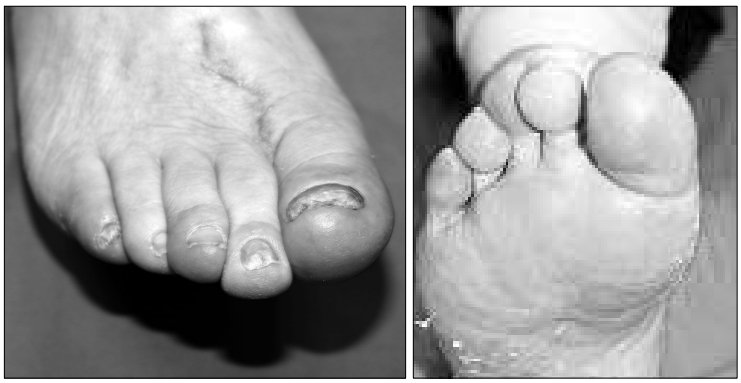

Fig. 5. After two months the patient had no clinically demonstrable problems.

closed with 10-0 nylon interrupted sutures (Fig. 4). The patient was not treated by anticoagulation and antiplatelet agents.

On follow-up examination 3 months postoperatively, symptoms were disappeared and he had no clinically demonstrable problems (Fig. 5).

III. DISCUSSION

Blue toe syndrome is occurred by the dislodged atheroemboli from atherosclerotic plaques in the aorta and arteries of the lower extremities, with subsequent occlusion of small vessels leading to ischemia, necrosis, organ damage and death.

Patient with blue toe syndrome usually experiences a painful discoloration of the toes. Unlike acute arterial occlusion, blue toe syndrome has normal pedal pulses. While acute arterial occlusion typically produces pallor, blue toe syndrome often produces cyanosis and livedo reticularis. The combination of toe cyanosis and preserved pedel pulses strongly implies atheromatous embolization and is often referred to as blue toe syndrome.[2] A common finding of blue toe syndrome may affect only a single toe, but usually involves several toes.[3] Our patient was also involved the 3rd and 5th toe as well as 1st toe. Cyanosis and livedo reticularis occur when the cutaneous venous plexus becomes visible because of increased amounts of desaturated venous blood or because of venodilation. It arises when atheroemboli obstruct small arteries, sluggish blood flow into the venous plexus and causing the blood there to stagnate and deoxygenate.[3]

The clinical manifestations of blue toe syndrome are variable depending on the size and location of the atheromatous plaque. When the plaque is located in the proximal aorta, the central nervous system, the abdominal viscera, and all four of the extremities can be involved. If it is located below the renal artery, only the lower extremities are involved.[3] Our patient presents with blue toe syndrome of microemboli to distal arterioles of the toes, originating from lower abdominal aorta or common iliac artery.

Spontaneous developed blue toe syndrome is rare.[4] It is more commonly occurred as an iatrogenic cause, such as diagnostic angiography and angioplasty.[4,5] Also, heparinization, thrombolytic therapy and surgical operations, especially aortic surgery, can cause blue toe syndrome.[3,6] Recently, the incidence of iatrogenic atheroemboli reveals a gradual increase because of extending indications for vascular surgery or invasive percutaneous procedures.[4,5] Our case was perhaps related to the angio CT performed to the diagnotic procedure.

Some commonly abnormal laboratory findings may contribute to diagnosis. It is reported that erythrocyte sedimentation rate is elevated in almost all patients.[2,3] Our patient was elevated level of erythrocyte sedimentation rate and c-reactive protein. Also, blood eosinophilia occurs in about 60% to 80% of cases,[3] but, our case is normal.

To confirm the diagnosis of blue toe syndrome, skin biopsy are often nonspecific and a biopsy of the gastrocnemius or quadriceps muscle may be required.[6] Although there was no histologic confirmation in our patient, the clinical findings strongly favored the diagnosis because the triad of foot pain, livedo reticularis, and intact pedal pulses is pathognomonic.

Currently, there are no specific treatments for blue toe syndrome and debates as to how best treat. Most authors recommand the removal of the atheroembolic source to treat effectively,[2,3] Because patient presents with a stenotic lesion that promotes production of microemboli, treatment the obstructing lesion looks like logic. Actually intra-arterial stents have proved successful in treating obstruction and theoretically supply a matrix that would

prevent plaque embolization.[3] However, there is an opportunity for producing additional emboli during stent placement procedure. Ohki et al. reported that significant emboli produced during the placement of stent into carotid endarterectomy specimens.[8] Another controversial issue is the use of anticoagulation. Many reports showed the causative role of anticoagulation in producing microembolization.[1-3] Acute ischemic extremity is most commonly the result of arterial occlusion secondary to thrombosis or embolization. Management of acute ischemic extremity is aimed at restoration of blood flow using anticoagulants. But, anticoagulation therapy in patient with blue toe syndrome can actually worsen the situation, presumably by inhibiting endothelialization of ulcerated atherosclerotic lesions making it promote to embolization.[3] Because microsurgical atheroembolectomy don't cause additional atheroemboli, we effectively treat our patient.

Diagnotic imaging tools of blue toe syndrome may be included a venous duplex to detect deep venous thrombosis, computed tomographic angiogram to reveal the source of atheromatous emboli, and echocardiogram to discover vegetations from endocarditis.[3]

A critical checkpoint in determining blue toe syndrome is a history, which should particularly elicit information about several issues, a recent invasive procedure involving the heart or a peripheral arterial vessel, the recent therapy of anticoagulant, previous arterial or venous thromboses. There is high risk of limb loss and high mortality rate in patients who present with blue toe syndrome. Physicians should be alert in the accurate diagnosis and follow-up to minimize adverse outcomes.

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