Case Report / Olgu Sunumu

Central Thromboangiitis Obliterans: A Case Report

Santral Tromboanjiitis Obliterans: Olgu Sunumu

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Abstract

A 47-year-old woman had inability to speak and swallow that had started 4-5 hours ago. She had necrotic lesions in the toes of both feet and had a history of smoking. Acute and chronic ischemic lesions were diagnosed on imaging. She was diagnosed with thromboangiitis obliterans by digital subtraction angiography. This report discusses a rare cause of ischemic stroke and the lack of data on diagnosis and treatment.

Key Words: Buerger's disease, Vasculitis, Stroke, Smoke, Neuroimmunology

Öz

47 yaşındaki kadında 4-5 saat önce başlayan konuşma ve yutkunma sorunu vardı. Her iki ayak parmak uçlarında nekrotik lezyonlar mevcuttu ve sigara kullanma öyküsü vardı. Görüntülemede akut ve kronik iskemik lezyonlar tespit edildi. Dijital substraksiyon anjiyografisi ile kendisine tromboanjitis obliterans tanısı konuldu. Bu raporda iskemik inmenin nadir görülen bir nedeni ve tanı ve tedavisine ilişkin veri eksikliği tartışılmaktadır.

Anahtar Kelimeler: Buerger hastalığı, Vaskülit, Inme, Sigara, Nöroimmünoloji

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Introduction

Thromboangiitis obliterans (Buerger disease) is an inflammatory disease characterized by non-atherosclerotic involvement of small and medium-sized vessels of the extremities (1). Buerger disease was first described by von Winiwarter in 1879 and named after Leo Buerger in 1908, who published the studies he conducted on amputated limbs of affected patients. The pathological feature of the disease is the presence of inflammatory thrombi in affected vessels; patients usually present with acute or chronic, ischemic or infectious acral lesions (2). Cerebrovascular complications are seldom observed (3,4). Herein, a case of thromboangiitis obliterans with central involvement was reported.

Case Report

A 47-year-old woman presented to emergency department with inability to speak and swallow that had started 4-5 hours ago. At physical examination she had a blood pressure reading of 130/70 mmHg, a pulse rate of 82 bom, a respiratory rate of 12/minute, and an oxygen saturation of 98%. She was conscious, had light reflexes +/+, isocoric pupils, normal eye movements, motor aphasia, muscle strength of 5/5 in all extremities, and a positive babinski sign in the right lower extremity. She had necrotic lesions in the toes of both feet (Figure 1). She had a history of smoking for ten years more than 10 cigarettes/day but no history of a chronic disease. Her blood tests revealed normal findings except for an elevated C-reactive protein level (49 g/l).

Figure 1. Necrotic lesions in toes

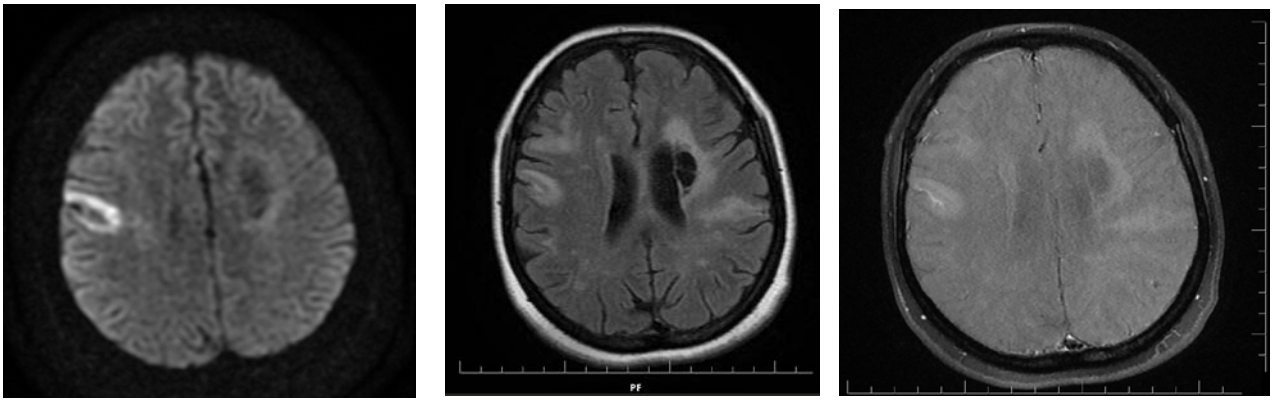

Figure 2. MRI; diffusion weighted image, T2 flair, contrast enhanced image.

A cranial diffusion weighted MR (magnetic resonance) study performed in the emergency department showed gyral restricted diffusion in the right parietal region as well as hyperintense lesions with a central cystic appearance in the left periventricular and left occipital regions (Figure 2). In a cranial MR imaging study with contrast, which was performed after the patient's admission to our clinic, these lesions were found to show contrast uptake (Figure 2). MR imaging venography showed no abnormality. A computerized tomographic angiography with contrast showed no abnormality in the intracranial and extra-cranial portions of the carotid and cerebral arteries. Tests for syphilis, human immunodeficiency virus, hepatitis, borrelia, FANA, rheumatoid factor, antineutrophil cytoplasmic antibodies, fluorescent antinuclear antibody,serum homocysteine, anticardiolipin antibodies and lupus anticoagulant, beta-2-glycoprotein I antibodies were negative. Levels of coagulation-re lated factors including the protein C, protein S and antithrombin III were all within normal limits. Additionally, a cardiac evaluation to look for any cardiac stroke etiology was also normal. A cerebrospinal fluid examination was planned but the patient refused it. She then underwent digital subtraction angiography, which revealed patchy ectatic changes in distal arterial branches in the cerebral region, particularly on the right side; additionally, a non-homogeneous staining was noted in distal parenchyma, which was interpreted in favor of vasculitis . Because of the similar appearance in the distal part of the lower extremities, thromboangiitis obliterans was considered as the primary diagnosis. The patient was advised to perform a vascular biopsy, but she did not accept it. The patient stopped smoking on our recommendation.

The department of cardiovascular surgery started oral cilostazol after iloprost infusion. Her speech and swallowing difficulties were attributed to pseudobulbar palsy; she was started on pyridostigmine, which improved difficulty swallowing at her follow-up.

Discussion

Thromboangiitis obliterans (TAO) often involves small and medium-sized vessels. Its prevalence is approximately 5-12 per 100,000 population each year in the world (5). Heavy tobacco use is prominent in the etiology, but it is multifactorial (6). The clinical diagnostic criteria include age under 50 years, a positive smoking history, presence of ischemia of lower extremities and typical arteriographical signs, and exclusion of diseases causing atherosclerosis, prothrombic disorders, and autoimmune disorders (7). It has been reported that approximately 15% of patients have cerebrovascular involvement before peripheral vascular involvement, and that both peripheral and cerebral symptoms last up to 20 years (8). There are cases reporting involvement of the cerebral arteries, coronary arteries, intestinal arteries, and aorta (9). Our patient presented with cerebral ischemic symptoms and silent infarctions and fulfilled all these clinical criteria of TAO.

Vascular histopathology provides the definitive diagnosis. Pathological findings such as worm-like occluded vessels which are not usually seen in atherosclerosis, can be characteristically seen at least in some patients with central TAO (3). Our patient did not accept surgical biopsy. In previous studies, cerebral angiographic findings have shown occlusions in the internal carotid arteries and intracranial major vessels, but these are not specific to TAO also be seen in atherosclerosis (3). The typical angiographic signs include the corkscrew appearance and abrupt interruptions in arteries (6). As in this patient, angiographic signs are also visible on cerebral vascular structures; however, they cannot be differentiated from primary cerebral vasculitis. In isolated central nervous system angiitis, multifocal arterial occlusions and collateral formation may be observed in association with segmental narrowing and dilatations (10,11). Therefore, its sensitivity and specificity in diagnosis are unknown. There is no definitive treatment for central TAO. As for treatment of peripheral vascular pathology, there are studies indicating that stopping smoking and other tobacco products significantly reduces amputation rate (1). A significant improvement was observed in the lesions on the toes in our patient. However, no study has yet reported on the central effect of this treatment. The absence of ischemic infarction episodes even though we did not give immunosuppressive treatment after quitting smoking, supports us that it can be effective in both diagnosis and treatment. Antiplatelet therapy may be useful to prevent white thrombus occlusions in leptomeningeal vessels, but supporting data are lacking (12). If there is an association with conditions that increase clotting, anticoagulant treatment is recommended.

We think that the small number of cases in the literature is a possible reason for our limited awareness of this disease (3,12,13). Furthermore, no study has yet investigated how these patients should be followed up, and there is a need for studies in this direction.

Ethical Approval: *On 26.10.2023, written informed consent was obtained from the patient.*

Author Contributions:
Concept: A.G
Literature Review: A.G
Design : A.G
Data acquisition: A.G,V.K
Analysis and interpretation: A.G
Writing manuscript: A.G
Critical revision of manuscript:A.G

Conflict of Interest: *The authors have no conflicts of interest to declare.*

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