



Case report

LOW BACK PAIN: ALWAYS A NEUROLOGICAL PROBLEM? A CASE OF LERICHE SYNDROME IN A WOMAN

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ABSTRACT

Leriche syndrome is defined as a rare obliteration of the aortic bifurcation. It is characterized by a typical triad in male patients: claudication, erectile dysfunction and decreased distal pulses. The leading cause of this syndrome is atherosclerosis. Possible differential diagnoses include vascular and neurological diseases. We report a case of a 54-year-old woman whose main complaint was low back pain. The initial (wrong) diagnosis was spinal disc herniation, causing neuropathy. The use of computerized tomography led to the correct diagnosis: aortoiliac occlusive disease (Leriche Syndrome).

KEYWORDS: *Leriche syndrome, atherosclerosis, aortoiliac occlusive disease, arteriopathy, low back pain, neuropathic pain*

INTRODUCTION

Leriche syndrome, also known as Leriche disease or carrefour disease or aortoiliac occlusive disease, is a condition that affects the blood supply to the lower extremities, typically the legs. It is defined as an obstructive chronic peripheral arteriopathy. The obstruction is usually found at the level of the iliac bifurcation (1). It is a rare pathology; its incidence is 1 in 12000 patients with aortic occlusions in an autopsy study (2).

It is caused by the occlusion or blockage of the main blood vessels leading to the legs, including the aorta and iliac arteries. This results in the typical symptomatology in male patients: claudication, leg pain, impotence, decreased pulse in the legs and lower extremities pallor. In severe cases, it can cause gangrene, needing amputation (3). However, sexual

Received: 04 January 2023

Accepted: 10 February 2023

ISSN: 2038-4106

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dysfunction such as hypoactive sexual desire disorder, dyspareunia and vaginismus can also affect women; this is due to the obstruction of blood flow due to atherosclerosis involving the iliac vessels causes a reduction in vascular flow at the vulval, vaginal and clitoral levels. Another explanation for reduced sexual function in women with Leriche Syndrome is ischemic damage of the pudendal nerve responsible for the innervation of the genital organs (4). Again, the incidence is higher among male individuals. However, this disease may also interest women, even if at a more advanced age compared to men (5, 6).

This syndrome was described for the first time by Robert Graham in 1914. However, in 1940, French surgeon René Leriche (Fig. 1) described a syndrome of thrombotic obliteration of the aortic bifurcation. He also described the typical triad of symptoms: claudication, erectile dysfunction and decreased distal pulses (7, 8). The first cause of this pathology is the deposition of atherosclerotic plaque in the aortic bifurcation; rare causes may be the presence of emboli and vasculitis (9). Like all arteriopathies, Leriche syndrome also has other risk factors: hypertension, diabetes mellitus, dyslipidemia, coronary artery disease, atrial fibrillation, primary anti-phospholipid antibody syndrome, hemodialysis and smoking (10, 11).

The differential diagnosis includes other vascular diseases such as abdominal aortic dissection or neurological pathologies such as peripheral neuropathy, vertebral stenosis, spinal disc herniation and Guillain-Barré syndrome (12). Diagnosis is based on symptoms, measuring the ankle-brachial index and imaging. In particular, colour doppler ultrasonography generally shows a reduction or absence of flow in the lower limbs. However, computed tomographic angiography is considered the gold standard for diagnosis (13). The most modern techniques allow the identification of the atheromatous plaque (14).

In the past, the first choice of treatment for the aortoiliac disease was aortofemoral bypass surgery with excellent results: patient survival ranging from 64 to 95% at 5 years (15). However, more recently, revascularization with an endovascular method gained more importance, and it is now considered first-line therapy. This technique is associated with significantly lower peri-operative morbidity and mortality rates than bypass (13, 16). Moreover, the success rate of endovascular therapy ranges from 73% to 100% (11, 17, 18). Also, medical and pharmacological approaches to prevent the progression of Leriche syndrome exist. They target the main risk factors of atherosclerosis: hypertension, hyperglycemia, hyperlipidemia and homocysteine. In addition, the use of antiaggregant therapy such as aspirin is fundamental. Another extremely important behavioural change is quitting smoking (10, 19). In addition, a physiotherapy program has been shown to improve walking ability and reduce leg pain by 50% - 200% (20).



Fig. 1. René Leriche (1879–1955)

CASE REPORT

This case report is of a 54-year-old post-menopausal woman with a previous medical history of anxiety-depressive syndrome untreated. She reported smoking about 20 cigarettes daily and being moderately active (amateur runner). She also reported having maternal familiarity with atherosclerosis. However, in the last year, she started complaining of low back pain with associated dysesthesias in both thighs, and she also reported constant cramping pains in her calves, preventing her from running. In addition, the patient complained of dyspareunia and vaginismus.

For this reason, she was referred to her General Practitioner (GP) and treated with Thiocolchicoside and Ibuprofen. However, her GP sent the patient for an orthopaedic evaluation when symptomatology persisted. The orthopaedic, suspecting a possible disc herniation, prescribed her Magnetic Resonance Imaging (MRI) of the lumbar spine. The imaging investigation showed minimal protrusion in the left median paramedian site of the L3-L4 disc and at the level of L5-S1, the presence of an inter-somatic disc with signs of degeneration, reduction in height and the presence of minimal hernial focus below the right paramedian median ligament that imprints the ventral surface of the dural sac (Fig. 2). After an evaluation of the patient



Fig. 2. RM sagittal: little herniated disc in L5-S1 (arrow)

and the imaging results, the orthopaedic prescribed her Oxycodone hydrochloride with Paracetamol and Ketoprofen.

The patient was prescribed physiotherapy, therapy with Etoricoxib 60 mg for 10 days and the possible use of a semi-rigid orthopaedic corset. However, despite the therapy, the woman continued to complain of the same symptoms. For this reason, she carried out a neurological examination. The neurologist prescribed electromyography, which documented chronic neurogenic suffering in the L4-L5 radicular competence area. Eventually, the patient carried out an ozone therapy session. However, after an initial benefit, the symptoms persisted. After evaluating the discrepancy between the symptomatology and the imaging, the neuroradiologist prescribed a computed tomography (CT) control. The CT scan documented the presence of a fibrocalcific atheromatous plaque at the level of the aortic bifurcation.

In light of the CT scan results, the neuroradiologist suggested she investigate more with imaging techniques, more specifically abdominal ultrasound and doppler ultrasonography of aorta and iliac vessels (Fig. 3). The Abdominal Ultrasound also confirmed the presence of atheromatous plaque involving 60-65% of the aortic lumen.

Only at this point, a precise diagnosis of Leriche syndrome is formulated. Consequently, the patient performed a vascular surgical visit in which she was advised to make a CT-angiographic study, suspend the habit of cigarette smoking, and start therapy with an antiplatelet agent (aspirin). However, she refused to perform a CT-angiographic study, and no clear information about her smoking habit is present.

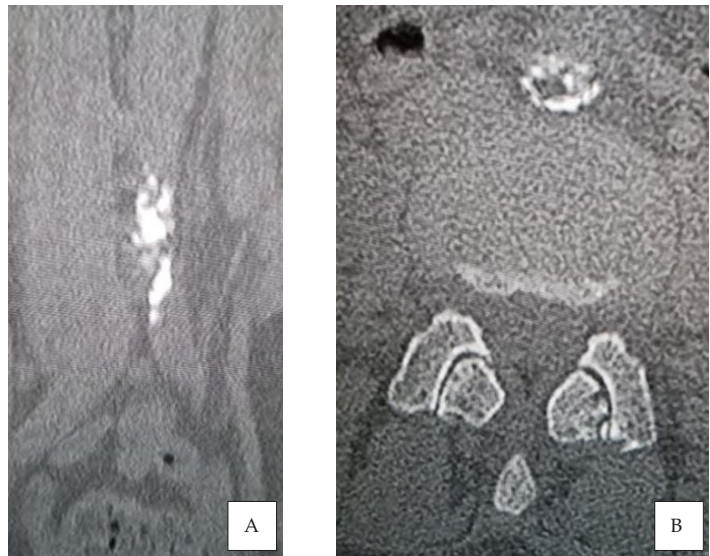


Fig. 3. CT scan of fibrocalcified atheromatous plaque at aortic bifurcation level with a 60-65% stenosis: A) coronal view, B) axial view.

DISCUSSION

As it is known, Leriche syndrome is an abdominal aorta or bilateral iliac occlusive disease that is caused by atherosclerosis (21). This syndrome has a slow progression, which provides the development of collateral circles; therefore, symptoms are nonspecific and insidious (22). The most common symptoms include claudication, low back pain, decreased leg pulse, pallor of lower extremities and sexual dysfunction (3, 4). The last symptom is more common in men (25-39%) but can also involve female patients (4). The possible origin of these symptoms may be neurological, but all other causes need to be excluded. A deep and precise imaging and clinical study are needed in specific cases. This report highlights that the key to the correct diagnosis in this pathology was reached only thanks to an advanced imaging methods such as CT and doppler ultrasonography (23, 24). This case report shows a rare case of Leriche syndrome: first of all, the patient is female and therefore, the typical symptomatological triad is absent. Moreover, the anxious-depressive syndrome of the patient led to a delayed diagnosis.

CONCLUSIONS

Lower limb weakness and claudication are neurological symptoms that may be present in Leriche syndrome. However, they can be caused by other factors such as cerebrovascular injury, spinal disc herniation, peripheral nerve disease, neuromuscular junction disease, muscle pathology, or metabolic conditions (25-29). Therefore, in case of doubtful neurological symptoms refractory to pain-relieving medical therapy, positive anamnesis and presence of risk factors for atherosclerosis, presence of sexual disorders not due to pathologies of the genital organs, it is always necessary to perform a diagnostic deepening. For example, as in this case, CT was performed to evaluate the vascular compartment and the

possible presence of atheromatic plaques. In conclusion, in the case of bilateral and symmetrical symptomatology, non-typical spinal herniation and other non-neurological problems must be investigated.

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