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A "Way Too Effective" Compensation with in the Leriche Syndrome. A Case Report

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List of Abbreviations:

ABI: Ankle-Brachial Index; CT: Computed Tomography; CIA: Common Iliac Artery; DSA: Digital Subtraction Angiography; EIA: External Iliac Artery; LS: Leriche Syndrome; MDCTA: Multidetector Computed Tomography-Angiography; MRI: Magnetic Resonance; PAOD: Peripheral Arterial Occlusive Disease; PTA: Percutaneous Angioplasty; TASC: Trans-Atlantic Inter-Society Consensus Document on Management of Peripheral Arterial Disease

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1. Abstract

Digital subtraction angiography (DSA) provides relevant information within the study and the treatment of Leriche Syndrome. In fact, besides confirming computed tomography (CT) findings (such as quantification of stenoses/occlusions and evaluation of collateral circulation), DSA offers a "dynamic" imaging of the vascular anatomy and pathology, including in-flow and distal runoff.

We hereby describe the case of a 53-year-old man with complete occlusion of the left common iliac artery and moderate stenosis of the contralateral iliac artery; notably, on CT examination, opacification below the level of the occlusion on the left lower limb appears faster and more dense than on the right, with nearly symmetrical clinical findings. Digital subtraction angiography examination before stenting demonstrates hypertrophic lumbar and hypogastric collaterals, the site of a "right-to-left" theft.

Multi-modal evaluation of Leriche Syndrome provides important information about the physiopathology of the disease, explaining unexpected clinical aspects.

2. Introduction

In Leriche Syndrome, diagnosis is straightforward and based on pathognomonic clinical and imaging features. Risk factors like smoking, hypertension, dyslipidaemia, and buttock and thigh claudication should be immediately addressed through proper diagnosis, later confirmed by contrast-enhanced CT or MRI. However, despite their high accuracy and reproducibility, both modalities provide "static" imaging of vascular anatomy and pathology. On the other hand, digital subtraction angiography before endovascular treatment (percutaneous angioplasty/PTA or stenting) provides the highest imaging quality and additional information about occlusions/stenoses, like in-flow and distal runoff.

3. Materials and Methods

A 53-year-old man with a history of hypertension, hypercholesterolemia, and heavy smoking is admitted to our hospital due to bilateral claudication of the buttocks and thighs (especially on the left). Foot pulses and ABI (Ankle-Brachial Index) are diminished (especially on the left: 0.45); on Doppler ultrasound examination, flow within the left common iliac artery is undetected.

4. Results

Contrast-enhanced CT demonstrates complete occlusion of the left common femoral artery and moderate (50%) stenosis (due to a calcified plaque) within the contralateral artery and mixed atheromatous plaques throughout the infra-renal aorta/bifurcation; notably, opacification below the level of the occlusion/stenosis appears earlier and more dense on the left lower limb (Figure 1).

DSA confirms occlusion of the left CIA (common iliac artery) and stenosis of the right CIA. It demonstrates marked hypertrophy of the lower lumbar arteries (especially on the left side) and inflow within both internal iliac arteries and their branches (Figure 2).

"Kissing" stenting is successfully performed with immediate improvement of bilateral iliac inflow and symptom relief (Figure 3).

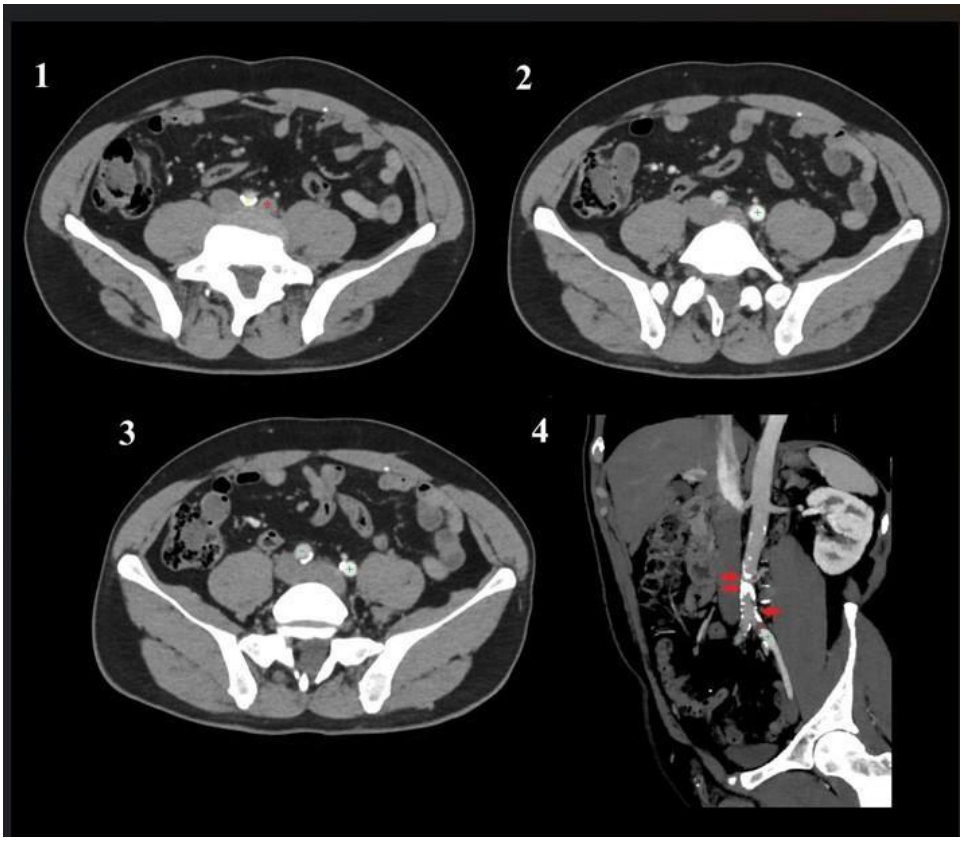

Figure 1: sequential axial scans (Figures 1-3) and multi-planar oblique reconstruction (Figure 4) from a contrast-enhanced CT. Segmentary (8 mm-long) thrombotic occlusion at the origin of the left common artery is seen (red asterisks), as well as calcific stenosis (of about 50% of the lumen) of the right common artery (yellow asterisk) and many calcific plaques throughout the infra-renal aorta (red arrows). Notably, despite the different degrees of the disease, contrast enhancement below the level of the lesions mentioned above appears more intense on the left lower limb (green plus) than on the other side (green minus).

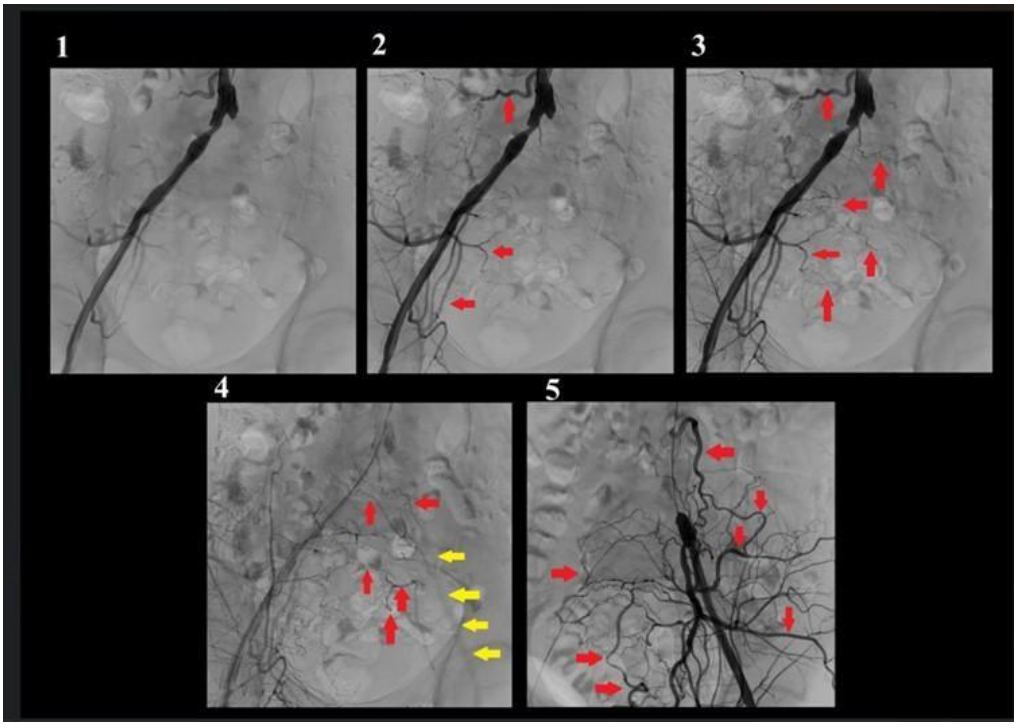

Figure 2: sequential frames in AP-views from aortoiliac DSA before treatment. After bilateral common femoral artery access (6-Fr sheaths) is obtained, aortoiliac DSA from a right-sided 5-Fr "Pig" catheter is performed, confirming complete occlusion of the left CIA and moderate stenosis of the right CIA and progressively showing bilaterally hypertrophic lower lumbar and iliac collaterals (**red arrows**) allowing distal run-off at the level of the left external iliac artery (**yellow arrows; Figures 1-4**). DSA from the left 6-Fr sheath better demonstrates the above-mentioned hypertrophic collaterals (**red arrows; Figure 5**).

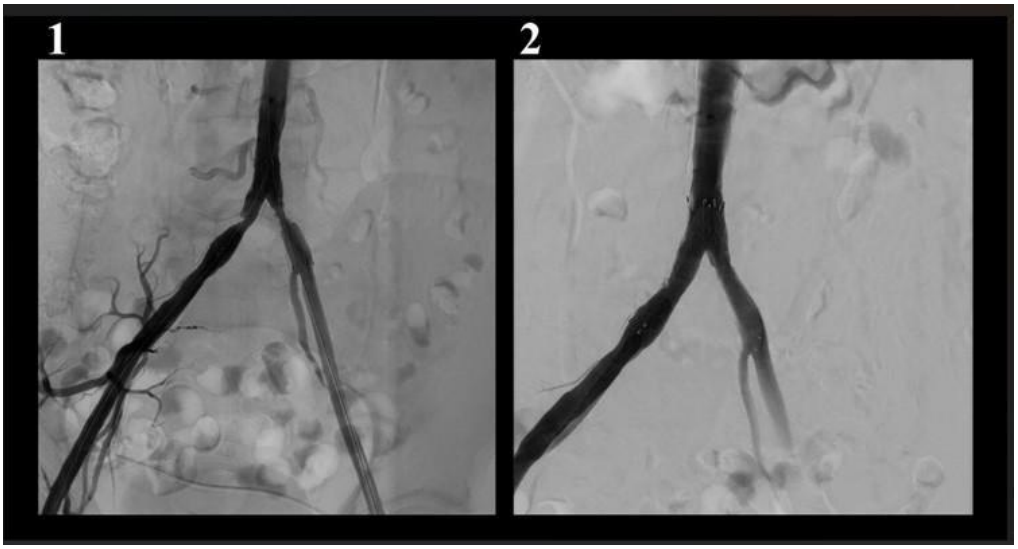

Figure 3: sequential frames in AP-views from aortoiliac DSA following the primary release of two nitinol self-expandable stents (9x60 mm) with "kissing technique" (Figure 1) and subsequent PTA with balloons (7 mm; Figure 2); an evident improvement of the aortoiliac inflow is seen.

5. Discussion

Leriche Syndrome (LS) represents a serious complication of atherosclerosis that affects the distal abdominal aorta, the iliac arteries, and the femora-popliteal vessels [1].

It usually affects subjects around 50 years [2] with atherosclerotic diffuse steno-obstructive disease and a history of smoking, hypertension, diabetes, and dyslipidaemia [3].

The atherosclerotic lesions are usually found at the aortoiliac level with four “patterns”: a) isolated stenosis of the common iliac arteries, b) more or less extensive lesions of the aortic bifurcation involving the origin of the common iliac arteries, c) extensive lesions of the abdominal aorta and iliac arteries (as in our case), and d) complete occlusion of the infra-renal aorta [2].

The main symptom is claudication, a cramping leg pain reproducible by exercise and relieved by rest. When bilateral common iliac artery involvement occurs, buttock claudication and even erectile dysfunction may be associated. Weakened or absent femoral pulses are found, with a decreased ankle/brachial index (ABI; normal values 0.9–1.3, claudication <0.50, severe PAOD 0.49–0.20).

Doppler ultrasound examination often represents the primary imaging modality for screening since it is non-invasive, inexpensive, reliable, and reproducible, although dependent on the operator, machine, and patient.

To date, multidetector computed tomography-angiography (MDCTA) is the most available and commonly used modality for diagnosis and evaluation of aortoiliac occlusive disease, allowing precise evaluation of the stenotic arterial segments, quantification of the occlusion, extent assessment, determination of collateral circulation, demonstration of pathological findings around the vessels, and planning of interventional/surgical approaches [4].

Digital subtraction angiography (DSA) historically represents the ‘‘gold standard’’ in PAOD, given the high quality of the study as well as the “dynamic” imaging of the vascular anatomy and pathology (including in-flow and distal runoff); however, it is invasive and expensive [5].

The prognosis of LS without treatment is usually poor; in some cases, with slow progression, collaterals may develop as a self-compensating mechanism [4].

Transluminal angioplasty is now advancing over surgical bypassing, sometimes representing the first-line treatment [2].

In particular, the endovascular approach is advisable as the first-line strategy in all TASC A through C aortoiliac lesions, given the low morbidity and mortality rates when compared with surgery, the high technical success, and good results in terms of patency, limb salvage, and survival rates, with lower costs and hospital stay and without the preclusion of any surgical option [4].

Nitinol self-expandable stents represent the most diffusely and frequently employed stents, thanks to the high conformability along curved segments [6,7].

Whenever the lesion involves the aortic bifurcation, the double-balloon/stenting technique is performed; in particular, primary stenting is advocated with more complex iliac lesions and occlusions and prevents distal embolization by fixation of the atherosclerotic/thrombotic material to the vessel wall [8].

Our patient presents many of the typical risk factors associated with LS (hypertension, hypercholesterolemia, and smoking above all). Despite the progressive nature of the disease (which has allowed abundant collateral formation), symptoms’ worsening has occurred with sudden, complete thrombosis of the left CIA. Due to the extent of the atheromatous lesions (unilateral CIA occlusion, referred to as TASC B), the patient is amenable to endovascular treatment. Dynamic high-quality imaging with DSA demonstrates both hypertrophy of the lower lumbar arteries (more evident on the left side) and marked in-flow within the inner branches of the internal iliac arteries, allowing distal

run-off within the left EIA. Moreover, the higher opacification detected on CT downstream on the left side finds an explanation in a right-to-left shunt/"theft" through the internal iliac arteries' branches (with the retrograde flow on the left side); this may cause the appearance of LS symptoms also on the right side, despite the non-critical degree of the stenosis. Primary stenting with the kissing technique is chosen to avoid distal embolization (given the mixed composition of the atheromatous plaques) and cover the contralateral CIA as well as the lowest segment of the infra-renal aorta.

5. Conclusions

Multi-modal evaluation of Leriche Syndrome not only allows proper diagnosis and planning of interventional/surgical procedures but also offers "functional" information about the physiopathology of the disease, especially in its clinical aspects. While CT allows diagnosis and assessment of the severity of the disease, only DSA can fully establish in-flow and distal run-off in case of an occluded vessel, sometimes demonstrating "paradoxical" compensations.

6. Declarations

All procedures performed in the study followed the ethical standards of the institutional and/or national research committee and the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

The patient provided written informed consent for using anonymized data for publication.

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

The authors declare that they have no competing interests.

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