Aortitis and Aortic Occlusion in Crohn Disease

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Patients with Crohn disease (CD) or ulcerative colitis are known to be at increased risk of arterial thromboembolic complications. We report the case of a 33-year-old woman suffering from CD for 19 years who presented lower limb claudication. Computed tomography scan revealed an aortoiliac occlusion extending from the level of the inferior mesenteric artery to both iliac bifurcations. Endovascular recanalization was attempted as a first option but failed. We then performed an aortobi-femoral bypass through a left retroperitoneal approach that allowed a total relief of the symptoms. Histologic study of the aorta demonstrated a nonspecific aortitis with lymphohistiocytic cell infiltration in the media and adventitia tunica. There was no signs of associated vasculitis. At the light of a literature review, we discussed our surgical strategy and the inflammation of the aortic wall as local factor of thrombosis that has never been previously described.

Inflammatory bowel diseases (IBDs) are characterized by gastrointestinal tract chronic inflammation. Most common vascular complications in IBD are venous thromboembolic events.

Arterial thrombotic events such as mesenteric, cardiac, cerebral arteries occlusions,[1–3] or mural thrombosis of the abdominal aorta[2,4–6] have been also described.

Our case is the only case of aortic occlusion associated to IBD for which histologic analysis of the aortic wall was performed. Histologic analysis showed a nonspecific aortic wall inflammation. Thereby, local inflammatory factors leading to aortitis and playing a role in arterial thrombus formation can be discussed in IBD patients.

CASE REPORT

A 33-year-old woman was referred to our institution in 2012 for a disabling bilateral lower limb claudication that occurred 3 months before.

She was suffering from Crohn disease (CD) since 1994 with several acute outbreaks that required intravenous steroid therapy in 1995, 1996, and 1997, and that has been complicated with rectovaginal fistula and perianal abscesses surgically treated in 2002. She received between 1994 and 2002 several cures of steroid therapy associated with azathioprine (75 mg up to 150 mg). Since 2002, the disease was stable, without any hospitalization or acute outbreaks under a treatment associating azathioprine (150 mg per day) and infliximab (300 mg per 8 weeks).

Her only cardiovascular risk factor was a 10 pack-year tobacco addiction. She did not take any hormonal contraception and had no history of deep venous thrombosis.

On 2012, she consulted for a fast-worsening claudication of both the thighs for a 20 m walking distance. No pulses were found on both the limbs.

Duplex scan demonstrated a distal aortic thrombosis extending to both proximal common iliac arteries (CIAs). Lower limb arteries were free of atherosclerotic

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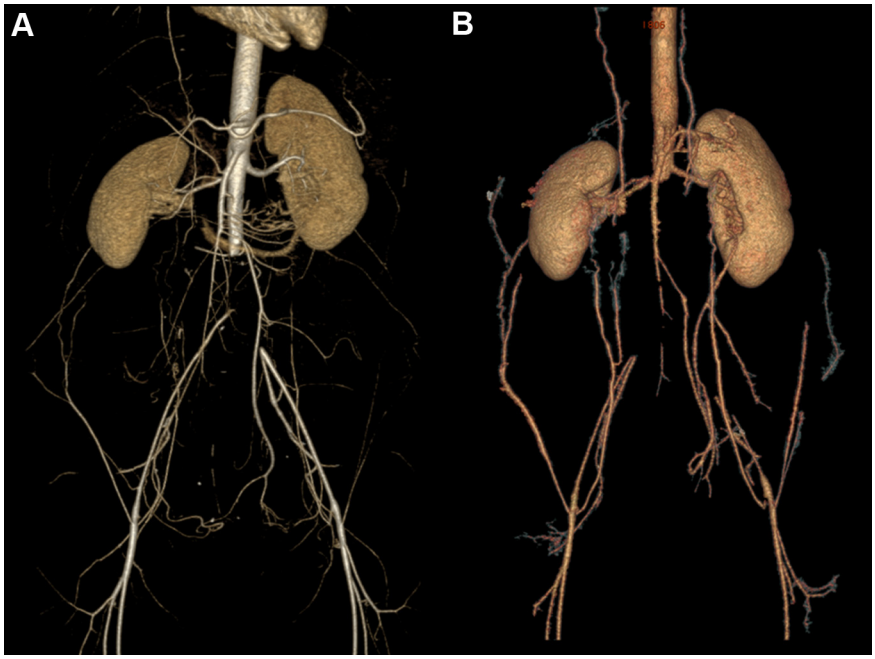

Fig. 1. (A) CT scan performed on May 2012 demonstrating the distal abdominal aorta occlusion starting below the inferior mesenteric artery and extending to both proximal common iliac arteries. **(B)** CT scan performed on November 2012 showing the proximal extension of the occlusion to just below the right renal artery.

lesion, but distal flow was weakened. Angio computed tomography (CT) scan showed an occlusion of the distal abdominal aorta starting below the inferior mesenteric artery and extending to both proximal CIAs without extension to the visceral arteries (Fig. 1A).

Biology excluded an associated thrombophilia: prothrombin time >70%, activated cephalin time = 1, fibrinogen <4 g/L, protein C <70%, protein S >65%, antithrombin III >80%, no factor V Leiden mutation and G20210 A prothrombin gene mutation, homocysteinemia <13.9 μmol/L, and no antiphospholipid antibodies.

Electrocardiogram was normal and echocardiography did not find any argument for cardiac embolism.

We first unsuccessfully attempted an endovascular treatment. The patient refused open surgery but came back 5 months later because of the persistence of disabling claudication. A new angio CT scan showed a proximal extension of the occlusion up to the renal arteries (Fig. 1B).

An aortobi-femoral bypass with a polyethylene terephthalate graft was performed in 2013 through a left retroperitoneal approach. After a 150 UI/kg intravenous injection of heparin, the aorta was clamped between the left and the right renal arteries to perform a safe thrombectomy of the juxtarenal aorta and a proximal end-to-end anastomosis just below the renal arteries. End-to-side distal anastomoses were performed on both common femoral arteries (CFAs). Peroperatively, the aortic wall did not look atherosclerotic, but very inflammatory. Arterial diameters were 11 mm for the abdominal aorta, less than 4 mm for CIA, and 5 mm for the CFA.

Postoperative course was uneventful, and she was discharged on day 7 with 1 month of preventive anticoagulation (enoxaparin, 4,000 UI/day) and lifetime antiplatelet therapy (aspirin, 75 mg).

Histologic examination of an infrarenal aortic specimen showed a nonspecific aortic wall inflammation with lymphohistiocytic cell infiltration of the media associated with adventitial inflammation (Fig. 2). The nature of the inflammatory infiltrate is confirmed by immunohistochemistry with the common leukocyte marker (clone 2B11 + PD7/26, DAKO) and CD163 (clone 10D6, MICROM). The aortic lumen was occluded by a fibrinocruoric thrombus.

There was no argument for Takayasu disease either on ultrasound examination or on histologic examination.

At 3-month follow-up examination, the patient did not suffer anymore from claudication and all pulses on both limbs were found.

A colonoscopy was performed in May 2013 to reassess CD activity. It found ulcers and multiple polyps with signs of active chronic inflammation on histologic examination. This aspect of a residual evolutive condition of CD associated to the histologic signs of aortitis led us to propose increasing the doses of immunosuppressive therapy: infliximab increased to 400 mg per 8 weeks with the same dose of 150 mg of azathioprine per day.

DISCUSSION

Thromboembolic events are well-known extra-intestinal complications of IBD. Patients with CD