Clinical Case Report

AORTIC THROMBOSIS – A SINGULAR MANIFESTATION OF CROHN DISEASE

TROMBOSE AÓRTICA – UMA MANIFESTAÇÃO SINGULAR DA DOENÇA DE CROHN

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ABSTRACT

Aortic thrombosis (AT) is an unusual extraintestinal manifestation of Crohn's disease (CD). We present a case of a male submitted to supracondylar amputation of the right lower limb due to irreversible acute ischemia. During the post-operative period, he developed signs of contralateral limb ischemia and performed complementary diagnostic tests which revealed a thrombus in the aortic bifurcation, a terminal ileum thickening, compatible with CD, and a polypoid lesion in the distal rectum. After patient staging and colorectal surgery, the complete resolution of AT was demonstrated.

Keywords: *Crohn disease; thrombosis; aorta, abdominal; rectal neoplasms.*

RESUMO

A trombose aórtica (TA) é uma manifestação extraintestinal rara da doença de Crohn (DC). Reporta-se um caso de um doente submetido a amputação supra-condiliana do membro inferior direito por isquémia aguda irreversível. No período pós-operatório desenvolveu sinais de isquémia do membro inferior contralateral, tendo realizado vários exames complementares de diagnóstico que revelaram a presença de um trombo na bifurcação aórtica, um espessamento do íleo terminal, compatível com DC, e uma lesão polipóide no reto distal. Após estadiamento do doente e realização de cirurgia colorretal, verificou-se a resolução completa do TA.

Palavras-Chave: *doença de Crohn; trombose; aorta, abdominal; neoplasias retais.*

INTRODUCTION

Acute limb ischemia (ALI) is an increasingly common condition in emergency department due to an aging population.[1] Despite thrombosis being one of the main causes of ALI, the presence of thrombus in the aorta in unusual. Aortic thrombosis (AT) is a rare disorder with high morbidity and mortality. It is more common in patients with atherosclerosis and may be related to hypercoagulable state, such as

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cancer, inherited thrombophilia and inflammatory bowel disease (IBD).[2,3] IBD is a heterogeneous group of diseases which involve gastrointestinal tract as well as other organs and systems.[4] An extremely rare and less studied complication of Crohn's disease (CD) is AT.[5]

The aim of this case is to report acute arterial thrombosis as a singular manifestation of IBD and incidental discovery of CRC, although IBD is related to colorectal cancer (CRC) via many factors.

CASE REPORT

A 50-year-old male went to the emergency department with a cold and cyanotic right lower limb (RLL) for 4 days. He was a smoker and a regular alcohol consumer. Physical examination suggested irreversible acute limb ischemia (grade III in Rutherford classification). He was submitted to a supracondylar amputation and in the post-operative period he developed mottled cyanosis which evolved into focal necrosis on the left toes, suggestive of atheroembolism.

The patient had an abdominal computed tomography angiogram (CTA) that revealed a partial eccentric thrombosis in the abdominal aorta, involving the common iliac arteries, and a concentric and diffuse thickening of the terminal ileum (figure 1). He also performed a colonoscopy with biopsies that described an infiltrative and eroded mucosa, which conditioned cecal lumen obstruction, and a sessile and friable polypoid lesion in the distal rectum.

The treatment was influenced by the patient's symptoms. The patient started anticoagulation with low molecular-weight heparin (LMWH) in therapeutic doses after amputation of RLL and was submitted to laparoscopic ileocecal resection due to intestinal obstruction initiated during the study. At the same operative time, transanal minimally invasive surgery (TAMIS) was performed to excise rectal lesion.

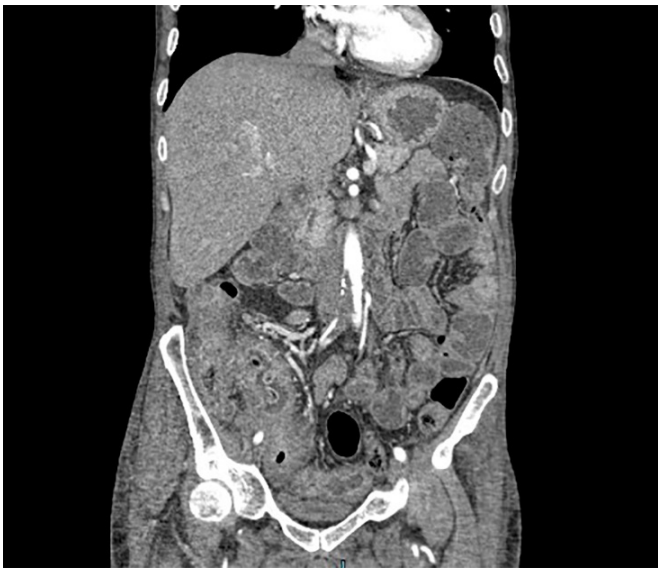

Figure 1 – **Abdominal CT-angiography shows a thrombus in the aortic bifurcation and thickening of the terminal ileum.**

After surgery he had an abdominal CTA that revealed a complete resolution of AT (figure 2).

The patient was discharged on 15th day post-surgery under anticoagulation with LMWH.

Histological analysis concluded that the changes in the terminal ileum were compatible with

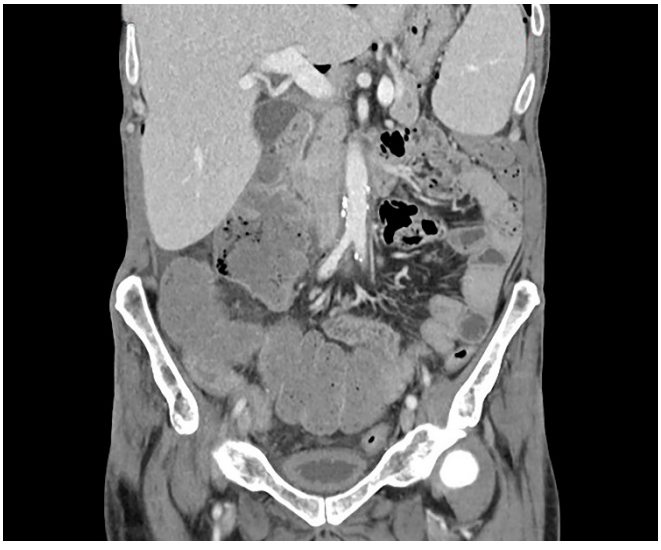

Figure 2 – **Abdominal CT shows complete resolution of aortic thrombosis.**

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CD and confirmed the presence of low-grade adenocarcinoma (G2) We achieved a R0 resection and the lesion was classified as pT2 Nx. After discussion in the multidisciplinary group, a total mesorectal excision (TME) was considered by the multidisciplinary group but rejected due to its comorbidities and lack of family support.

After discharge, he maintained regular follow-up at surgery and vascular surgery appointments.

DISCUSSION

Early diagnosis of AT is difficult because most patients are asymptomatic and it depends on imaging tools available (CTA or MRI). However, ALI can be the first manifestation of AT.[6] Usually the study of colorectal diseases, such as CD and CRC, is performed in patients with suggestive symptoms.[4] In this case, the path was slightly different. The first manifestation of CD was an AT leading to embolic complications of both lower limbs, which warranted other complementary diagnostic tests, and allowed its diagnosis.

AT found in abdominal CTA is rare in CD and few cases are described in literature. It is more frequent in younger patients and occurs mainly in distal abdominal aorta extending to the iliac arteries.[5] These patients have higher rate of complications and overall poor outcomes.[7] The mechanism of AT in IBD is not completely understood. A history of smoking, the usage of hormones, the activity and extension of disease, postoperative status and genetic factors have been reported as risk factors.[7,9] Thus, patients with severe active CD have more inflammatory mediators in circulation, which promotes a prothrombotic state.[9,10] In our patient, smoking is the only factor identified, whose relationship is well established.[4,10]

On the other hand, IBD has been associated with CRC for several years. IBD leads to chronic inflammation of the intestinal mucosa, which favours dysplasia and carcinogenesis. Although dysplasia is a predisposing factor there are few cases of CRC derived from isolated adenomatous lesions, which arise in areas of normal mucosa.[11,12]

The management of these patients is challenging and they benefit from a multidisciplinary team composed by gastroenterologists, vascular and colorectal surgeons.[13] The treatment should be individualized according to the severity of CD and patient comorbidities. Therapeutic strategies involve treatment of underlying IBD and prevention of acquired risk factors.[6] The first therapy line of CD includes multiple drugs. Surgery treatment must be well thought out and applied when medical treatment fails.[14]

Different approaches have been proposed for AT. Medical therapy with anticoagulants is effective and can be responsible for complete resolution of thrombus without surgery. Surgical strategies include endovascular or open approaches and must be performed in AT recurrence.[6] Our patient started anticoagulation therapy instead of aortic procedure, because complete resolution of AT was achieved after ileocecal resection, which allowed a decrease of inflammatory process and anti-fibrinolytic activity.[6,13]

In conclusion, AT related to IBD is an uncommon condition with serious complications for which clinicians should be aware. The best treatment is still a matter of debate and regular follow up must be performed.

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