



LOWER EXTREMITIES GANGRENE IN PATIENT WITH ULCERATIVE COLITIS

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ARTICLE INFO

Article History:

Received 11th January, 2018
Received in revised form
18th February, 2018
Accepted 29th March, 2018
Published online 30th April, 2018

Key Words:

Extremities gangrene,
Thromboembolism, Ulcerative colitis,
IBS, Reperfusion injury,
Coagulopathy.

ABSTRACT

Objective: Increased coagulability is a rare but well-recognized feature which complicates Inflammatory Bowel Disease. Thromboembolism associated with ulcerative colitis has been reported since 1936 but there is no absolute therapeutic management protocol for thromboembolic complications yet.

Case presentation: A 14-year-old girl was brought to our emergency department due to pain and paresthesia in both lower extremities and history of bloody diarrhea for 40 days before admission. Because of bloody diarrhea she underwent colonoscopy with biopsy the results were in favor of Ulcerative colitis. Pulses of both lower extremities were absent. Color Doppler showed thrombosis of both iliofemoral arteries from infrarenal aorta with intact veins.

Discussion: Thrombosis in arteries can usually be treated by thrombectomy, anticoagulation and compartment fasciotomy in cases of limb ischemia.

It has rarely been reported thrombosis causing bilateral limb gangrene resulting in amputation. In this case due to no response to medical and surgical treatment right above and left below the knees amputation was performed with colectomy to control disease and eventually discharged after closure of ileostomy patient continued warfarin therapy.

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Citation: Zeraatian, S., Mesbah, M., Kamalzadeh, N., Hosseini, M., Naseripour, M., Pazooki, D., 2018. "Lower extremities gangrene in patient with ulcerative colitis", *International Journal of Development Research*, 8, (04), 19896-19898.

INTRODUCTION

Thromboembolism associated with ulcerative colitis has been reported since 1936 (Bargen, 1936) Arterial and venous thromboses have been seen in 1.2%-6.4% of patients. Thrombosis in arteries can usually be treated by thrombectomy, anticoagulation and compartment fasciotomy in cases of limb ischemia, has rarely been reported Thrombosis causing bilateral limb gangrene resulting in amputation. Because of its rarity, a classic approach has never been documented in this situation. Here, to remind the possibility of aforementioned situation and possible therapeutic pathway we present our case.

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Case

A 14-year-old girl was brought to our emergency department due to pain and paresthesia in both lower extremities and history of bloody diarrhea for 40 days before admission. No history of fever, weight loss, skin lesion or other medical problems was reported. On physical examination femoral artery pulses just beneath, inguinal ligaments in both lower extremities were absent with coldness and mild discoloration of skin, especially below the knees but upper extremity pulses were all intact. Color Doppler showed thrombosis of both iliofemoral arteries from infrarenal aorta with intact veins. Impression of arterial thrombosis was made, heparin started and bilateral thrombectomy was performed via an inguinal incision. She developed skin bolus in her left foot and after eight hours, signs and symptoms of ischemia in both extremities represented.

The patient was re-assessed with Doppler which showed re-thrombosis despite full anticoagulation and steroid pulse therapy. This viscous cycle of re-thrombosis was repeated several times and finally ended in gangrene of both lower extremity below knees region. After repeated thromboembolectomy, patient also showed evidence of reperfusion syndrome including hyperkalemia and thrombocytopenia (Fig-1).

Because of bloody diarrhea she underwent colonoscopy with biopsy the results were in favor of Ulcerative colitis, so she received *sulfasalazine* (1.5gr /d), *methylprednisolone* (10mg/kg) and *cyclophosphamide*. Her abdominal sonographic assessment and echocardiography were normal. The patient did not have any significant changes in liver function test, protein C, protein S, antithrombinIII, anticardiolipin, factor V and VIII, eventually due to failure of drug therapy in management of colitis and thrombosis, she underwent restorative proctocolectomy and ileoanal anastomosis with proximal diversion ileostomy. Plasmapheresis also was performed postoperatively for stopping the viscous cycle of thrombosis and to save lower extremities. But unfortunately, re-thrombosis finally resulted in gangrene of both lower extremities. Ultimately right above and left below the knees amputation was performed with closure of ileostomy and patient was discharged on warfarin.



Figure 1. Gangrene of below knee region

DISCUSSION

Increased coagulability is a rare but well-recognized feature which complicating Inflammatory bowel disease (Bargen, 1936). Thrombotic complications are usually the result of pro-coagulant changes in the blood of patients with IBD (especially ulcerative colitis). Arterial thrombosis is another rare complication with a reported incidence of less than 1 in 1000 patients with IBD. The vessels noted to be involved include the aortoiliac, femoro-popliteal, and digital arteries (Jackson, 1997). It generally occurs as a postoperative complication and has been found to be more common in *Crohn's disease* than ulcerative colitis. In different case reports and reviews preferential involvement of lower extremity by thrombosis in inflammatory bowel disease were showed (Talbot, 1986; Suarez Crespo *et al.*, 1997). Thrombosis involving multiple sites is an extremely rare feature in Inflammatory bowel disease. *Jackson et al* reported 5 out of 52 patients with ulcerative colitis who had thrombosis and

thrombotic episodes involving two different sites or at different occasions (Jackson, 1997). Three of them had three different thrombotic events. They occur in young adults whose disease is active. Good clinical improvement was achieved after treatment with steroids, sulfasalazine and anticoagulation with enoxaparin, thrombectomy followed by long-term warfarin. Concomitant use of drugs, such as sulfasalazine and azathioprine, for ulcerative colitis has been shown to result in warfarin resistance (Teefy *et al.*, 2001).

In addition to efforts to restore the blood flow as effective means in sudden and refractory thrombosis of the arteries, resection of the large bowel can be an option. Restorative proctocolectomy with *ileal pouch-anal anastomosis* (IPAA) is the surgical treatment for ulcerative colitis but in acute phases subtotal colectomy suffices however, surgery is not without danger and complication, and colectomy in ulcerative colitis results in either a life- long ileostomy or the need for further surgery to create a reservoir (Weiss *et al.*, 1995). For these reasons, medical therapy should be used to control the disease and allow the patient to retain the colon if the patients have developed no severe complications. Colectomy should be considered in patients who continue to deteriorate, or do not improve during a one- to two-week period. Intravenous cyclosporine (4 mg/kg per day) or oral micro-emulsion cyclosporine, tacrolimus has been used as alternative to colectomy (Treem, 1991).

Conclusion

Arterial thrombosis in IBD setting can cause end organ damage, so reperfusion should be the priority in the treatment. Thrombectomy or thrombolysis which are accompanied by systemic anti-coagulation are the priority in the treatment of patients with UC and arterial thrombosis. Surgery should be considered as the last resort in some circumstances in which the re-thrombosis endangers the end organs, besides supplementary para-clinical tests which should rule out other treatable hypercoagulable states.

Disclosure: The authors declare no conflicts of interest.

Acknowledgement

Many thanks to Z Hosseini PhD student, for checking and statistics.

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