



SHORT REPORT

Fournier's gangrene complicating ulcerative pancolitis

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Abstract

Fournier gangrene is a very rare and a rapidly progressing, polymicrobial necrotizing fasciitis or myonecrosis of the perineal, perianal and genital regions, with a high mortality rate. Infection is associated with superficial trauma, urological and colorectal diseases and operations. The most commonly found bacteria are *Escherichia coli* followed by *Bacteroides* and streptococcal species. Diabetes mellitus, alcoholism, and immunosuppression are perpetuating co-factors. Fournier's gangrene complicating inflammatory bowel disease has been reported in three patients so far, two with Crohn's disease.

A 78-year-old man diagnosed with ulcerative pancolitis was referred for fever, and painful perianal and scrotal swelling after perianal surgery for a horseshoe-type perianal abscess. Since bowel disease diagnosis, patient was on mesalazine and achieved long-term remission. Perianal abscess occurred suddenly one week before perianal surgery without any evidence of pre-existing fistula or other abnormalities. Physical examination showed extensive edema and crepitus of perineum and genitalia and patient had symptoms of significant toxicity.

The diagnosis of Fournier's gangrene was made and patient underwent emergency surgery with extensive surgical debridement of the scrotal and perianal area and Hartman procedure with a diverting colostomy. In addition, patient started on therapy with mesalazine 3gr, methylprednisolone 16 mg, parenteral nutrition and broad spectrum of antibiotics. Two days after the first operation the patient needed a second operation for perianal debridement. On the fourth day, blood cultures showed *E. coli*. Patient had an uneventful recovery and was discharged after 34 days of hospitalization. On follow up, disease review is scheduled and colostomy closure is planned.

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1. Introduction

Fournier gangrene is a rapidly progressing, polymicrobial necrotizing fasciitis or myonecrosis of the perineal, perianal and genital regions,¹ with a mortality rate ranging from 15% to 50%.^{2–4}

Jean Alfred Fournier, a French venereologist in 1883, first described Fournier gangrene. The cause is a polymicrobial infection associated with superficial traum,^{5–7} urological diseases and operations^{8,9} and colorectal diseases including carcinoma,¹ diverticulitis,¹⁰ retroperitoneal appendix,¹¹ foreign bodies or staplerhemorrhoidectomy.¹²

In women, additional causes have included septic abortion, Bartholin gland or vulvar abscess, episiotomy, and hysterectomy. Insect bites, burns, trauma, and circumcision have been reported as causes of pediatric Fournier gangrene, which is rarely seen.¹

The most commonly found bacteria are *Escherichia coli* followed by *Bacteroides* and streptococcal species.¹³ Diabetes mellitus, alcoholism, immunosuppression and other severe chronic illnesses are frequent perpetuating co-factors.¹⁴

Fournier's gangrene is characterized by a sudden onset most commonly without prodromal symptoms.¹⁵ Early diagnosis and aggressive wide debridement, combined with early colostomy¹⁶—and cystostomy if needed—hemodynamic stabilization, analgesia¹⁷ and intravenous administration of broad spectrum antibiotics are the keys to successful treatment.¹⁸

Although the actual incidence of Fournier gangrene is unknown, the disease is relatively uncommon.

We present herein an exceptional case of a patient with long-standing ulcerative pancolitis who was diagnosed and successfully treated for Fournier's gangrene.

2. Case report

A 78-year-old man diagnosed with ulcerative pancolitis was referred to the Department of Emergencies of hospital exhibiting fever, perianal and scrotal swelling and pain.

Patient underwent elsewhere perianal surgery for a horseshoe-type perianal abscess 72 h prior and since then complained of perianal pain and fever. Patient had been diagnosed with ulcerative colitis twenty years ago. At that time and during follow up all endoscopies and subsequent bowel biopsies were compatible with ulcerative pancolitis and Crohn's disease or any other chronic bowel diseases were excluded. According to clinical examination and patient history there was no evidence of perianal disease. In addition, upper gastrointestinal endoscopy and subsequent histology were normal and terminal ileum appeared normal at all ileocolonoscopies with normal biopsies. Small bowel follow through and abdominal computed tomography were normal. No extraintestinal manifestations or significant comorbidities were so far recorded.

Since diagnosis, patient was started on mesalazine monotherapy and achieved long-term remission with 1.5 gr/d mesalazine maintenance treatment for the last ten years. For several years preceding this admission, patient had had neither bowel abnormalities nor other types of intestinal or extraintestinal complaints. The clinical course

of the disease during the period of the detection of the complex perianal fistula was mild. However, two months before admission patient complained of some abdominal cramps and slight urgency in defecation that was successfully managed with combined administration of metronidazole and ciprofloxacin. Perianal abscess occurred suddenly one week before perianal surgery without any evidence of pre-existing fistula or other bowel abnormalities.

Physical examination showed extensive edema and crepitus of perineum and genitalia (Fig. 1) with fat stranding and a foul-smelling discharge also present. Patient exhibited symptoms of significant toxicity but there was no abdominal distention and no discomfort or rebound. In detail, vital signs were as follows: blood pressure at 100/62 mm Hg, temperature at 39.6 °C; heart rate was 98 bpm and respirations at 17 per minute.

Arterial blood gases were normal with pH at 7.34. Laboratory investigation revealed elevated white blood cell count (23,200/mm³), thrombocytosis (576,000/mm³), with hemoglobin at 12.1 gr/dl, C-reactive protein at 290 mg/dl, while biochemical profile was unremarkable with no signs of organ failure.

The diagnosis of Fournier's gangrene was made with those clinical and laboratory criteria and patient underwent emergency surgery. Extensive surgical debridement of the scrotal and perianal area and a Hartman procedure with a diverting colostomy was performed (Fig. 2). In addition, patient started on therapy with mesalazine 3gr, methylprednisolone 16 mg, parenteral nutrition and broad spectrum of antibiotics. Two days after the first operation the patient needed a second operation for perianal debridement. On the fourth day, blood cultures showed *E. coli* and antibiotics were modified accordingly. Although an ulcerative colitis flare was very probable at that time and all precaution were taken, of note colorectum status during the period of gangrene treatment was in remission and there were no signs of bowel disease relapse. Operational endoscopy was not possible at the time of the emergent surgery; however, the experienced colorectal surgeon did not notice any significant bowel abnormality on bowel margins.

Patient had an uneventful recovery and was discharged after 34 days of hospitalization. On follow up a new endoscopy and disease review is scheduled in order to re-evaluate initial diagnosis and plan colostomy closure.



Figure 1 Extensive edema and crepitus of perineum and genitalia with fat stranding in a patient with ulcerative colitis diagnosed with Fournier's gangrene.

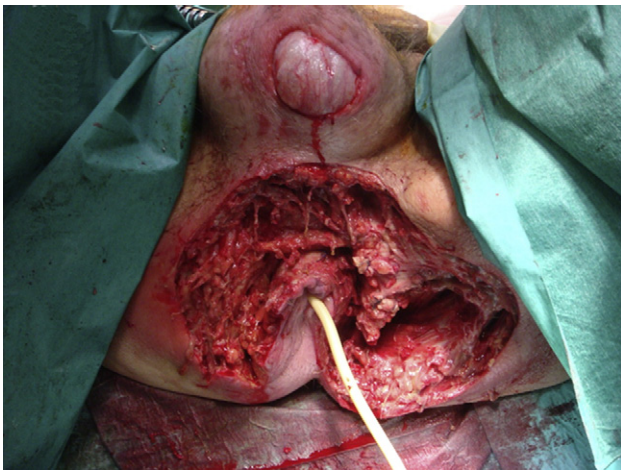


Figure 2 Extensive surgical debridement of the scrotal and perianal area followed by a Hartman procedure with a diverting colostomy in a patient with ulcerative colitis diagnosed with Fournier's gangrene.

3. Discussion

Fournier's gangrene complicating inflammatory bowel disease has been reported in three patients so far, two of them with Crohn's disease. In detail, reports include one patient with Crohn's ileocolitis¹⁹ one patient with previously undiagnosed Crohn's disease with a complex enterovesical fistula⁸ and finally one patient with ulcerative colitis, diabetes and alcoholism.¹⁶

In the patient with ulcerative colitis reported herein a surgical incision in the perianal area was the triggering factor; however, in many other cases the etiology leading to the spread of the infection remains unknown.

Fournier gangrene tends to be polymicrobial in nature, with synergy of aerobic and anaerobic bacteria. The most commonly found bacteria are *E. coli*—as in this patient herein—followed by *Bacteroides* and streptococcal species. The anatomic association between the facies of penis, scrotum, perineum, groin and gluteal favors the fast spread of tissue necrosis.²⁰ Fournier gangrene represents an emergency with a potentially high mortality rate. Prognostic factors include older age, duration of symptoms before hospitalization, urea and creatinine level on admission, associated systemic diseases, cancer diagnosis, the urological source of infection and the source, time of evolution and extent of necrotizing fasciitis.^{21,22} Of note, a Fournier Gangrene Severity Index assessing scoring before admission and at the time of discharge has been suggested.²³

The causes of death include severe infections, severe sepsis, multiple organ failure, coagulopathy, thrombosis, acute kidney failure and diabetic ketoacidosis.^{16,24,25}

This patient was operated twice within one week for his Fournier's gangrene. In fact, multiple debridement may be necessary to remove all nonviable tissue while hyperbaric oxygen has also been used as adjuvant treatment and may benefit patients who remain ill despite undergoing multiple or extensive debridements, although its role remains controversial.¹ According to the other three cases, Fournier gangrene in patients with inflammatory bowel disease was successfully

managed by debridement, broad spectrum antibiotics and hyperbaric oxygen.¹⁹ In Crohn's disease a diverting ileostomy was performed before skin grafting and scrotal reconstruction while bowel disease was treated with mesalamine, metronidazole, 6-mercaptopurine and infliximab.

As in this case, patient hospitalization exceeded one month. In fact, patients surviving Fournier's gangrene were hospitalized for an average of one month period while in the 5-year follow up only 50% of the patients were free of pain and in few sexual function was impaired.²⁶ Despite major complaints due to extensive scarring most patients consider their cosmetic result as well their quality of life to be satisfactory. Follow up of those patients is important as anal tumor occurring after Fournier gangrene has been reported.²⁷

To conclude, these are the particulars of a case of a patient with ulcerative colitis diagnosed and successfully treated for Fournier gangrene. When operated in the perianal area, in addition to Crohn's disease patients also patients with ulcerative colitis merit special attention and cautious management by experts in order to avoid unfavorable outcomes.

The importance of prompt diagnosis and intensive therapy of Fournier gangrene in specialized medical centers is of paramount importance in such exceptional cases.

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