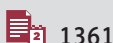


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Fournier's Gangrene During Pregnancy in a Patient with Crohn's Disease

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Corresponding Author: Lucas Faraco Sobrado, e-mail: fsobrado@gmail.com, lucas.sobrado@hc.fm.usp.br
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Final Diagnosis: Crohn's associated Perianal and perirectal abscess • Fournier's gangrene
Symptoms: Anal pain
Medication: —
Clinical Procedure: —
Specialty: Surgery**Objective:** Unusual clinical course**Background:** Fournier's gangrene (FG) is a rapidly progressive necrotizing infection of the perineum. Risk factors include male sex and immunosuppression. Inflammatory bowel disease and pregnancy may alter immune response by complex mechanisms but have rarely been associated with necrotizing infections of the perineum. To the best of our knowledge, only 5 cases of FG in association with IBD have been reported in the literature, and none of them occurred during pregnancy.**Case Report:** We report the case of a young woman with long-standing Crohn's disease in clinical remission with Infliximab monotherapy who developed FG in the third trimester of pregnancy. A cesarean section was undertaken at 35 weeks due to fetal distress, followed by debridement, diverting stoma, and vacuum-assisted therapy. The perineal defect was closed following 4 debridements and vacuum-therapy exchanges with a unilateral medial thigh advancement flap, and a draining seton was placed in the suprasphincteric fistula. The patient was discharged after 28 days and her recovery was unremarkable. The neonate also recovered well.**Conclusions:** The treatment of FG is multidisciplinary and includes early debridement and intestinal diversion. Perianal pain should not be disregarded, as it may be the initial symptom of severe perianal sepsis in the immunosuppressed. To the best of our knowledge, this is the first case report of FG during pregnancy in a patient with Crohn's disease.**Keywords:** Fournier Gangrene • Inflammatory Bowel Diseases • Negative-Pressure Wound Therapy • Pregnancy Complications • Reconstructive Surgical ProceduresFull-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/934942>

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Background

Fournier's gangrene (FG) was first described in 1883 by the French dermatologist Jean Alfred Fournier, who originally reported an idiopathic fulminant infection of the scrotum [1]. It is now recognized that immunosuppression associated with local trauma or urinary and colorectal infections are the most common etiologies [2]. FG has been rarely associated with inflammatory bowel disease (IBD) or pregnancy, although both conditions can affect the immune system [3,4]. The treatment of IBD, which includes immunosuppression and anti-TNF medications, can also interfere with the immune response and increase the risk for opportunistic infections [5]. Perianal disease is relatively common in IBD, particularly in Crohn's disease (CD), but rarely progresses to necrotizing infections. To the best of our knowledge, only 5 cases of FG in association with IBD have been reported in the literature, but none of them occurred during pregnancy [6-10].

We herein describe an uncommon clinical case of a young woman with long-standing CD in the third trimester of pregnancy who developed FG, which was successfully treated by a multidisciplinary team at our institution. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Case Report

We report the case of a 29-year-old woman in her 35th week of pregnancy. Her past history included Crohn's disease (CD) for 10 years involving the left colon and perianal fistula. Her IBD was in clinical remission with Infliximab monotherapy, which was continued through her pregnancy. Previously, she had a perianal abscess drainage 1 year ago and had failed Certolizumab Pegol. The patient had no other clinical comorbidities, HIV serology was negative, and prenatal care was unremarkable. She presented to the Obstetrics Department reporting mild perianal pain and was discharged following a reassuring clinical examination. Five days later, she returned to the Emergency Department with worsening pain and was admitted for fetal monitoring and colorectal evaluation. Physical examination was unremarkable except for an anal stricture. The initial work-up revealed a white blood cell count of 13 500 and C-reactive protein of 115 mg/L. A perineal ultrasound showed a 9-ml fluid collection. Intravenous ceftriaxone and metronidazole were started for the treatment of a small perianal abscess.

During the night the patient had to undergo an emergency cesarean section due to fetal distress, which was uneventful and the baby recovered well. In the following 12 h postpartum, she became toxic with tachycardia (HR 124 bpm) and hypotension (MAP 62 mmHg), the perianal pain progressed, and a



Figure 1. Fournier's gangrene with diffuse erythema (arrowhead), swelling of the vulva, and purple discoloration of the skin prior to debridement.



Video 1. Incision and drainage of Fournier's gangrene. Laparoscopic loop ileostomy showing enlarged uterus in the first postoperative day of cesarean section and intraoperative colonic lavage and final aspect following debridement and regional flap.

proctology examination revealed diffuse hyperemia with swelling of the vulva and purple discoloration of the skin (**Figure 1**). FG was suspected, and she was taken to the operating theater for wide drainage and debridement (**Video 1**). Intraoperative findings revealed necrosis of the ischioanal fat with foul-smelling purulent discharge that was debrided until visualization of healthy tissue (**Figure 2**). Anoscopy showed rectal ulcers and a fistulous opening with purulent discharge in the distal rectum. The anal sphincter was spared from necrosis. A laparoscopic loop ileostomy was undertaken with intraoperative colonic lavage through the distal limb. Vacuum-assisted therapy was performed and hydrocolloid paste was placed in the perineal defect. Cultures revealed polymicrobial infection with *Escherichia Coli*, *Citrobacter freundii* complex, and *Candida albicans*, and antibiotics were adjusted accordingly.



Figure 2. Final aspect following first drainage and debridement of necrotic tissue.



Figure 3. Final aspect of unilateral medial thigh advancement flap with a seton through the fistula tract.

The patient was discharged from the Intensive Care Unit after 3 days. On the 14th postoperative day, following 4 debridements and vacuum-therapy exchanges, the perineal defect was closed with a unilateral medial thigh advancement flap and a draining seton was placed in the suprasphincteric fistula (Figure 3). The patient was discharged after 28 days of hospitalization and her recovery was unremarkable. After 3 months of follow-up, the medial thigh flap was completely healed and showed no signs of infection. The seton was kept in place and Infliximab monotherapy was resumed. Ileostomy reversal is planned. The baby also recovered well and is on regular follow-up with the pediatrician.

Discussion

FG is a rapidly progressive necrotizing infection of the perineum, and failure to recognize this condition early has been associated with higher mortality [11].

Although early treatment is essential, the initial clinical picture may be misleading. Tissue necrosis with crepitus and discoloration of the skin are clinically evident only in the minority of patients [12]. During pregnancy, mild pain is common and can be unappreciated by the attending physician. In the present case, the initial assessment was unremarkable and the patient was discharged with painkillers but returned with worsening pain.

Perianal disease is common in CD and can affect up to one-third of patients [3], although it rarely progresses to severe necrotizing infection. To the best of our knowledge, only 5 cases of FG in association with IBD have been described: 2 in ulcerative colitis (UC) [7,9] and 3 in CD [6,8,10], but none of them during pregnancy. In detail, 1 patient had pancolitis from UC, diabetes and alcoholism [7]; 1 patient had long-standing UC and developed FG following perianal abscess drainage [9]; 1 patient had CD with enterovesical fistula [6]; 1 patient had undiagnosed and untreated CD ileocolitis [8], and 1 patient was a 62-year-old man with CD and type-2 diabetes [10].

Immunological changes in IBD are complex and may be related to the disease itself, malnourishment, or due to treatment. Our patient was on Infliximab monotherapy, which is an anti-TNF medication known to increase susceptibility to infections [5]. Immunological compromise plays a role in the development of FG, but it is more commonly associated with diabetes, HIV, alcoholism, and cancer [2].

The development of FG during pregnancy is also very rare and only a few cases have been reported in the literature, none related to IBD [13-16]. In the first 2 cases, severe infection caused premature birth and the newborn died within 48 h [13,14]. In 1 patient, emergency cesarean section and debridement were undertaken at the same time [15], whereas in the other, fulminant infection developed a few hours following birth [16], which was similar to our case.

In our patient, the primary source of infection was present before delivery and caused oligohydramnios, which was not present in the prenatal ultrasound and is a sign of fetal distress. Although pregnancy is a physiologic state, it has also been associated with increased risk for specific infections [4] and likely contributed to the development of FG.

The standard management of FG includes extensive debridement and antibiotics. The need for a diverting stoma is

controversial but has been advocated in cases of rectal perforation, fistulas, or fecal incontinence [17]. In the present case, the inciting event was clearly of colorectal origin, with perianal abscess and fistula, so we believe it was prudent to divert early to control sepsis and to facilitate management of vacuum-therapy. Since the patient had a diseased left colon from Crohn's colitis, our preferred option was a loop ileostomy with distal washout. A loop-transverse colostomy would be another option but is associated with higher risk of troublesome prolapse and parastomal hernias [18].

The management of vacuum-assisted therapy in the perineum is difficult due to loss of adherence and local contamination, leading to frequent dressing changes in the operating room. The use of hydrocolloid paste, such as demonstrated in **Figure 3**, can facilitate adhesion of the plastic film. Negative-pressure therapy has utility in controlling local sepsis and promoting tissue healing [19].

The resulting tissue loss from extensive debridement often leaves primary closure unfeasible. Reconstruction with regional flaps is safe and should be undertaken after all necrotic tissue has been debrided and healthy tissue is present [11]. Particular care should be taken in patients with CD due to impaired healing of the perineum in active disease. In our patient with perianal abscess and fistula, a seton was placed in the fistula tract through the flap to avoid recurrent abscess.

Conclusions

The treatment of FG during pregnancy is complex and involves a multidisciplinary team to ensure the care of the baby and to facilitate early debridement and intestinal diversion. Perianal pain should not be disregarded, as it may be the only symptom in an immunosuppressed patient.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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