

Fournier's gangrene in a patient with psoriatic arthritis: a case report

Gangrena de Fournier em paciente com artrite psoriática: relato de caso

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ABSTRACT

Background: Fournier's gangrene is a rare disease characterized by a perineal and abdominal necrotizing infection. The disease has an unfavorable prognosis without a prompt diagnosis and appropriate treatment. **Objective:** Present a Fournier's gangrene case in a post-bariatric patient in a methotrexate treatment for psoriatic arthritis. **Case presentation:** 58-year-old man was admitted with seven days of anal pain and unresponsiveness to antibiotic therapy. His past medical history included bariatric surgery and psoriatic arthritis, which was treated with weekly 17.5 mg doses of methotrexate. During the examination, he presented left lateral perianal hyperemia down to the scrotum and a painful digital rectal examination. Laboratory tests indicated an intense infectious process. Computerized tomography confirmed the laboratory findings. Images showed perineal and perirectal adipose tissue densification, suggesting Fournier's gangrene. The patient was submitted to perianal abscess drainage and debridement of necrotizing tissue. Perianal, gluteal, and left scrotum perineal regions were debrided. After that, the patient was referred to the ICU. Broad-spectrum antimicrobial therapy was administered. Clinical evolution was satisfactory, and the patient was discharged after 15 days. During home care, the patient developed pyelonephritis and COVID-19. Due to the patient's immune status, methotrexate treatment was discontinued, and acitretin was prescribed. **Conclusions:** Fournier's gangrene is a rare disease with an annual incidence of 1.6 cases per 100,000 males. Urogenital, anorectal, and genital skin infections in immunocompromised patients cause it. Early diagnosis is decisive for a good prognosis, considering the high mortality associated with the development of necrotizing fasciitis.

Keywords: Psoriatic arthritis; Gastric bypass; Methotrexate; Fournier's gangrene.

RESUMO

Background: Fournier's gangrene is a rare disease characterized by a perineal and abdominal necrotizing infection. The disease has an unfavorable prognosis without a prompt diagnosis and appropriate treatment. **Objective:** Present a Fournier's gangrene case in a post-bariatric patient in a methotrexate treatment for psoriatic arthritis. **Case presentation:** 58-year-old man was admitted with seven days of anal pain and unresponsiveness to antibiotic therapy. His past medical history included bariatric surgery and psoriatic arthritis, which was treated with weekly 17.5 mg doses of methotrexate. During the examination, he presented left lateral perianal hyperemia down to the scrotum and a painful digital rectal examination. Laboratory tests indicated an intense infectious process. Computerized tomography confirmed the laboratory findings. Images showed perineal and perirectal adipose tissue densification, suggesting Fournier's gangrene. The patient was submitted to perianal abscess drainage and debridement of necrotizing tissue. Perianal, gluteal, and left scrotum perineal regions were debrided. After that, the patient was referred to the ICU. Broad-spectrum antimicrobial therapy was administered. Clinical evolution was satisfactory, and the patient was discharged after 15 days. During home care, the patient developed pyelonephritis and COVID-19. Due to the patient's immune status, methotrexate treatment was discontinued, and acitretin was prescribed. **Conclusions:** Fournier's gangrene is a rare disease with an annual incidence of 1.6 cases per 100,000 males. Urogenital, anorectal, and genital skin infections in immunocompromised patients cause it. Early diagnosis is decisive for a good prognosis, considering the high mortality associated with the development of necrotizing fasciitis.

Descritores: Psoriatic arthritis; Gastric bypass; Methotrexate; Fournier's gangrene.

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INTRODUCTION

Fournier Syndrome is characterized by a necrotizing fasciitis of the perineum, genital and perineal region. The infectious process results in thrombosis of the subcutaneous blood vessels, which leads to gangrene of the overlying skin. All this is due to an obliterating endarteritis caused by the spread of microorganisms in the subcutaneous space⁽¹⁾. Although Jean Alfred Fournier first described it as an acute idiopathic gangrene in healthy young men⁽²⁾, this pattern has become an exception in modern medicine⁽³⁾. The leading causes are infections in the urogenital tract, anorectal area, and skin of the genitals⁽⁴⁾. Besides that, prevalence is higher in patients with pre-existing comorbidities such as diabetes mellitus, alcoholism, malignant neoplasms, cirrhosis, prolonged hospitalization, AIDS, lupus, and immunosuppressive chemotherapy⁽¹⁾.

Patients with psoriasis and psoriatic arthritis have an increased risk of infections. The most common infections to affect this population are in the respiratory tract, abdomen, skin, and soft tissues. The risk of complications increases with age, comorbidities, and history of infections⁽⁵⁾. Furthermore, immunosuppressive therapy with methotrexate frequently used in these pathologies is related to myelosuppression adverse effects⁽⁶⁾. Patients with a medical history of bariatric surgery bypass usually present malabsorption of the vitamin B complex, mainly of B12, after the procedure. B12 deficiency is known to suppress the immune function, increase the production of pro-inflammatory cytokines, and stimulate the NF- κ B⁽⁷⁾. With that in consideration, psoriasis and psoriatic arthritis may be related to Fournier's gangrene since they increase the risk of infections. The immunosuppression process associated with malabsorption of vitamin B complex and methotrexate drug therapy might be associated with the same reason.

We present the case of a 58-year-old man with previous bariatric surgery, in a methotrexate drug therapy for psoriatic arthritis, diagnosed with a severe form of Fournier's gangrene (FG). The patient provided written informed consent to publish his clinical information and images.

CASE PRESENTATION

A 58-year-old man with a medical history of psoriatic arthritis was treated with 17.5mg/week of methotrexate immunosuppressive therapy for six months. The patient also underwent a bariatric bypass surgery seven years ago. He was admitted to the emergency room at Hospital São Vicente (Guarapuava-Paraná, Brazil) with severe anal pain for seven days and a fever,

despite being on antibiotic therapy with ciprofloxacin. Antibiotic unresponsiveness and rectal abscess were hypothesized. In addition to the initial medical history, the patient also underwent cholecystectomy one year ago. Furthermore, he is a former smoker, 15 pack-years, with no comorbidities related to arterial hypertension or diabetes mellitus. Before admission, the patient self-medicated with ketorolac and dipyrone, showing temporary improvement in the symptomatology. The family reports that he resisted being taken to the emergency service, claiming he was fine. On physical examination, he presented with abdominal distension, tachycardia, sweating, left lateral perianal hyperemia up to the scrotum, and painful rectal examination.

Laboratory tests showed a hemoglobin level of 9.9g/dL, hematocrit at 27.6%, leukocytes at 35,240/mm³ showing vacuoles in neutrophils, platelets at 379,000/mm³, C-reactive protein at 28.64ng/dL, direct bilirubin at 1.71mg/dL and procalcitonin at 2.2ng/mL. Urine culture with colony count and blood culture results were negative. The patient then was submitted to an abdominal computerized tomography, which showed perineal and perirectal adipose tissue densification and blurring of the lower prostate's contours (Figure 1). Although the image showed no signs of impairment of the obturator internus muscles, the pattern described still is compatible with Fournier's syndrome.

The patient was submitted to perianal abscess drainage and debridement of necrotizing tissue. Perianal, gluteal, and left scrotum perineal regions were debrided. A left lateral incision was made, and drainage and debridement of the entire necrotic region down to the healthy tissue were performed. Necrosis was found



Figure 1. Computed Tomography of the abdomen shows densification of peri-rectal adipose tissue extending to the perineal region, suggesting an inflammatory/infectious process.

in the scrotum from the skin until the fat and cellulite to the left lateral periprostatic region. The internal anal sphincter was preserved. After that, hemostasis, cleaning with saline solution and chlorhexidine, bandage with neomycin, and compresses were performed (Figure 2). The patient was then referred to the ICU due to the risk of complications from the extensive injury and the possibility of septic shock.

The sample collected during surgery was submitted for anatomopathological analysis. The results showed intense acute inflammation and tissue necrosis compatible with necrotizing cellulitis.

The antimicrobial therapy prescribed was cefepime 2g, clindamycin 600mg, and neomycin 5mg/g + bacitracin 250ui/g. The patient had a satisfactory clinical evolution during the first five days in intensive care. During this time, therapeutic changes were made with the insertion of collagenase 0.6u/g + chloramphenicol 0.01g/g and metronidazole. Clindamycin and neomycin use was suspended. When transferred to isolation, the patient showed increased levels of leukocytes and a worsening general condition. Because of this, a new change in antimicrobial therapy was performed. This time, meropenem 2g IV and vancomycin 500mg IV were prescribed, and the patient's status showed significant improvement.



Figure 2. Immediate postoperative.

Clinical evolution was satisfactory, and the patient was discharged after 15 days. The patient was readmitted for one more period of hospitalization due to the developed pyelonephritis and COVID-19 during home care. Due to the patient's immune status, methotrexate treatment was discontinued, and acitretin was prescribed after consultation with the rheumatologist.

DISCUSSION

Fournier's gangrene is a rare disease with an annual incidence of 1.6 cases per 100,000 males. It is caused by urogenital, anorectal, and genital skin infections in immunocompromised patients with diabetes, obesity, and malignant neoplasms⁽⁴⁾. Symptomatology is associated with skin, subcutaneous tissue, and muscle necrosis. It can progress to sepsis, multiple organ failure, and death. In the laboratory examination, a general leukocytosis, or leukopenia, the appearance of young forms, and the toxic granularity of neutrophils are observed in the available blood test⁽⁴⁾. Radiography can also be used in the diagnosis of Fournier gangrene. Through the imaging, gas and liquid can be detected in the depth of soft tissues, which cannot be identified in the physical examination⁽⁴⁾.

Treatment of Fournier's gangrene involves emergency surgical intervention with antimicrobial therapy. Considering the polymicrobial character of fasciitis, broad-spectrum drugs that cover both anaerobic and aerobic bacteria are required. Prescription of 2nd and 3rd-generation cephalosporins with nitroimidazoles, fluoroquinolones, and aminoglycosides is recommended. In more complex cases, carbapenem association therapy can be used.

The only risk factor for the patient whose case we are describing presented was the immunosuppression caused by his autoimmune disease treated with a DMARD immunosuppressive therapy with an agent and by his previous bariatric surgery. No previous symptoms or blood parameters suggested the development of Fournier's gangrene. To the best of our knowledge, this is the first reported case of immunosuppression-related Fournier's gangrene due to psoriatic arthritis with immunosuppressive methotrexate therapy and bariatric surgery.

Psoriatic arthritis is a unique type of inflammatory arthritis associated with dermatological psoriasis and related to comorbidities that reduce the patient's quality of life⁽⁸⁾. A higher risk of serious infections was observed in a cohort study (Hazard ratio (HR) and (95% CI): 1.54 (1.44-1.65) in patients with severe psoriasis, regardless of the treatment used⁽⁹⁾. Considering that, the risk of

infections to which individuals with psoriasis and psoriatic arthritis may be exposed is evident.

Besides that, treatment of these pathologies with biological or conventional systemic agents may also be associated with different risks of serious infections. PSOLAR registry⁽¹⁰⁾ found that the risk of infection based on exposure within 91 days was: ustekinumab (0.89), infliximab/golimumab (2.86), and methotrexate (1.28) events per 100 patients observed. The most common serious infections were pneumonia and cellulitis. Thus, analyzing which immunosuppressive therapies may be more associated with infectious scenarios is possible.

Bariatric bypass surgery (RYGB) alters the gastrointestinal tract since it excludes parts of the small intestine, reducing nutrient absorption. Considering that the duodenum, jejunum, and ileum are involved in the absorption of vitamin B complex, the procedure can induce malabsorption of these vitamins, mainly B12 (7). Cobalamin deficiency can cause T cell proliferation and production of pro-inflammatory cytokines. It also increases TNF- α and affects the methylation reaction, increasing the inflammatory response and reducing CD8+ and natural killer cell activity. All ultimately lead to an immunosuppressive process.

Finally, this is the first reported case in the literature of immunosuppression-related Fournier's gangrene due to psoriatic arthritis with immunosuppressive methotrexate therapy and bariatric surgery in a patient with no other comorbidities commonly related to fasciitis. We emphasize the prompt diagnosis and a good prognosis, considering the high mortality associated with the development of Fournier's gangrene.

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DECLARATION OF COMPETING INTEREST

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

ETHICAL STATEMENT

The present article was conducted strictly with the Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving human subjects. The manuscript adheres to the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, aiming for the inclusion of representative human populations in terms of sex, age, and ethnicity, as per those recommendations. The terms "sex" and "gender" were used correctly. The authors explicitly state that informed consent was obtained from the patient. The privacy rights of the participants were rigorously observed throughout the research process.

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