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Fournier's Gangrene in a Man Who Was HIV-Positive With a High CD4 Count: An Unusual Presentation of a Complex Rectoscrotal Fistula

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ABSTRACT

Fournier's gangrene is a potentially life-threatening, necrotising fasciitis of the perineal and/or genital region. Known risk factors include diabetes, alcoholism, and immunosuppression. Often described as idiopathic, a cause can usually be found such as trauma, catheterization, or anorectal fistula. We report a case of Fournier's gangrene in a 52-year-old man who was HIV-positive but had an underlying complex fistula that was discovered during the course of his treatment. He was successfully treated with surgical intervention and antibiotics. This patient was not typical of most previously described cases because: (1) he had a low viral load and previously high CD4 count; (2) there was no initial evidence of traditional portals of entry for organisms; (3) there was no evidence of sexually-transmitted infection found in surgical specimens; and (4) the complex fistula was the likely nidus of infection.

INTRODUCTION

Fournier's gangrene (FG) is a form of necrotising fasciitis that affects the soft tissues of the genitalia [1]. The condition is named for Jean-Alfred Fournier, a French venereologist who described his findings in a group of young male patients at a lecture in 1883. However, Baurienne also described similar findings as early as 1764.

Although originally defined as an *idiopathic gangrene of the male genitalia* [1], a likely etiology can be found in 95% of cases [2]. These include anorectal and urogenital causes (eg, urinary tract infection, anorectal fistula). The perineum is often involved. Although the incidence is greater in men, both sexes can develop this form of gangrene.

Etiological factors such as trauma to the urogenital tract act as a portal of entry for microorganisms. Certain conditions including diabetes mellitus, immunodeficiency, and alcoholism are recognized factors leading to debility, hypoperfusion, and depressed cellular immunity [1,2]. Compromised immunity provides a favorable environment for initiation of infection, and the virulence of the microorganisms contributes to the rapid spread of the disease through the production of exotoxins and cytolytic enzymes. Tissue cultures usually grow a mixture of aerobic and anaerobic bacteria, typically Streptococcus sp. with other organisms including *Staphylococcus sp*., *Enterobacter sp*., and fungi [3].

KEYWORDS: Fournier's gangrene; Necrotising fasciitis; HIV/AIDS infection

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Abbreviations and Acronyms

AIDS = acquired immune deficiency syndrome
CD4 = cluster of differentiation 4
FG = Fournier's gangrene
HIV = human immunodeficiency virus

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case report

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FG is uncommon but not rare. It is estimated that there have been 500 cases reported since Fournier's original lecture in 1883, giving a prevalence of 1 in 7500 persons [1]. Other researchers performed a search using Medline (US National Library of Medicine) and other journals to estimate 600 cases reported between 1996 and 1998 [4]. The frequency of FG is not thought to have changed appreciably because there may have been a global increase in reporting of new cases.

The present case is unique because this infection developed in a patient who was otherwise healthy but positive for the human immunodeficiency virus (HIV). The case provides important lessons regarding management of the condition.

CASE REPORT

In 2004, a 52-year-old man presented to the emergency department with painful, swollen genitalia and purulent discharge from the dorsum of his penis. He was diagnosed HIV-1 seropositive in 1995. He began highly active antiretroviral therapy (HAART) in 1997.

Four weeks before his emergency-room evaluation, he had been treated with nitrofurantoin for a urinary tract infection by his general practitioner. However, his symptoms worsened with the development of fever and penile and scrotal cellulitis. He attended the sexual health clinic, where he was treated with coamoxiclav (a combination antibiotic containing amoxicillin trihydrate and potassium clavulanate) and an antihistamine.

No screening test for sexually transmitted infections (STI) had been performed in the previous 3 months, although the patient described protected receptive anal intercourse with casual male partners. He denied intravenous substance usage. His cluster of differentiation 4 (CD4) lymphocyte count at this time was 673 x 10^6/L. This count was within normal CD4 limits (500-1200 x 10^6/L), indicating that his disease was under control and he was not immune-deficient. His HIV-1 viral load (VL) was < 50 copies per mL. He was taking a combination of zidovudine, abacavir, and lamivudine. It should be noted that HIV infection causes a progressive reduction in the number of T cells expressing the glycoprotein *CD4* on their surfaces.

The patient's condition deteriorated further and he presented to the emergency room with a temperature of 37.5°C. His genitals were edematous, tender, and erythematous, and there was pus discharging from the penile shaft. Urinalysis was negative and routine blood tests were normal. On day 2 of his admission, urgent urological opinion was sought. This resulted in a clinical diagnosis of FG. He was taken to the operating room where the necrotic tissue was excised, revealing a fistula from his scrotum to the dorsum of the penis (Figure 1).

Figure 1. The Laid-Open Fistula of a Patient with Fournier's Gangrene.

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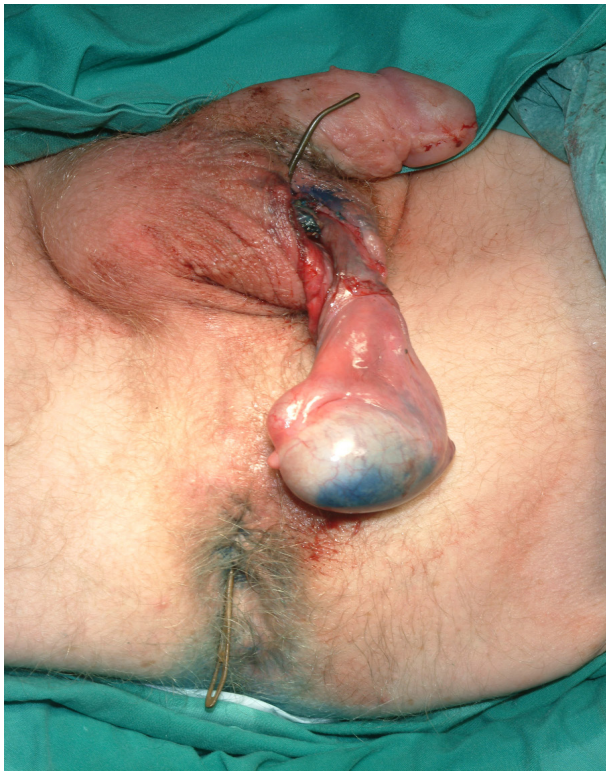

Note the probe (at the bottom of the wound) exiting from the left hemiscrotum (top). The left testis is viable.

Culture of the tissue grew mixed coliforms, with *Bacteroides sp*. predominating. He later underwent skin grafting to the penis and scrotum. Magnetic resonance image scanning was performed several months later because of persistent scrotal discharge (images unavailable); this revealed a second complex fistula involving the anus, rectum, and scrotum.

The patient was admitted electively, and the fistula was laid open by a general surgeon. The fistula was left to heal with a seton in situ.

The patient has since remained free of major HIV sequelae. In late 2009, he had a CD4 count of 914 (29%) and an undetectable viral load. He was taking a protease inhibitor-based combination. He still had mild penile deformity, lower urinary tract symptoms, and moderate erectile dysfunction. He is being managed successfully with tamsulosin and tadalafil.

DISCUSSION

The first case of FG in association with HIV or acquired immune

deficiency syndrome (AIDS) was documented in 1991 [5] in a patient with AIDS. Since 2001, only 11 similar cases have been reported in Western literature [6,7]. An epidemiological study in Zambia [8] demonstrated an increase in the incidence of FG during the start of the HIV epidemic in the early 1980s. The incidence rose from 1 case per year in the era before HIV to 10 cases in 1994, with 80% of these patients testing positive for HIV.

A few studies suggest a 50% increased risk of death from FG with every 3-fold rise in HIV viral load; however, others found that risk of death from FG relates to the patient's condition at presentation and delays in recognition and/or treatment rather than active underlying disease [9,10]. However, with aggressive therapy along established surgical lines, patients who are HIV-positive appear to recover as well as patients who are HIV-negative [8]. Although global reporting of FG has increased, the role of HIV infection in its etiology and natural course is still unclear.

Regardless of etiology, FG presentation is often variable. The onset may be insidious, as it was in our patient, with nonspecific features of malaise, fever, increasing pain, and swelling. Once necrotising infection has been initiated, the causative microorganisms typically produce toxins that cause thrombosis of nutrient vessels to the skin and subcutaneous fascia. The reduction in local blood supply results in a reduction in oxygen tension, allowing the rapid multiplication of facultative anaerobic and microaerophiliac bacteria and causing necrosis of the overlying skin. These organisms, in turn, produce lytic enzymes (eg, collagenases, lecithinases), which digest the fascial barriers and lead to rapid extension of the disease [11].

The combination of fascial digestion and necrosis is the hallmark of FG. It manifests as intensely painful, erythematous, swollen genitalia that may have visible areas of necrosis or a palpable *crepitus* caused by gas-forming microorganisms. Fulminant FG can involve the fascial envelope of the genitalia and the perineum and extend into the thighs (as far as the insertion of *fascia lata*). The systemic effects vary with the degree of necrosis, from low-grade pyrexia to florid septic shock [1,2,12].

Our patient was not typical of most previously described cases of FG in that: (1) he had a low viral load and previously high CD4 count; (2) there was no initial evidence of traditional portals of entry for organisms; (3) there was no evidence of sexually-transmitted infection found in surgical specimens (although formal anogenital samples were not taken); and (4) the complex fistula was the likely nidus of infection. However, it should be noted that this patient had no previous history of perianal or pelvic sepsis or trauma to that region; perianal sepsis is more prevalent in homosexual men with HIV infection (up to 4%) when compared with homosexual men without HIV (1.6%) [13].

The present case raises issues regarding the diagnosis and management of *all* patients with FG. Complete history-taking and examination are important to uncover risk factors and assess the extent of spread. Immunodeficiency is a risk factor, but it cannot be assumed to be the precipitant of FG. CD4 count and HIV viral load are important, but the mere presence of HIV infection does not establish a causal link to FG. Other associated factors such as a fistula should *always* be sought. In addition to blood grouping and typing, blood tests should include full blood count, coagulation screen, and renal biochemistry in order to evaluate any metabolic or hematological disturbances that may be sepsis-induced.

Imaging is of limited use and should not delay treatment, particularly in critically ill patients. Early surgical exploration and debridement of necrotic tissue with concomitant broad-spectrum antibiotic therapy restores organ perfusion, reverses organ failure, and reduces mortality [14,15]. Fecal or urinary diversion may be required if there is perineal involvement; early involvement of colorectal and reconstructive surgeons facilitates the recovery of these patients. Repeated exploration and excision is often required over a period of several days to ensure that all nonviable, infected tissue has been removed. In addition, treatment of the etiological factor (eg, fistula) should not be neglected if full resolution of sepsis is to occur.

Results following even extensive debridement are often excellent with reconstruction using split-skin grafts and/or local myocutaneous flaps. Up to 50% of men with penile involvement will have some degree of discomfort upon arousal after reconstruction. Most are satisfied with the postoperative cosmetic result, but psychological counseling may also be needed [15].

REFERENCES

1. Paty R, Smith AD. Gangrene and Fournier's gangrene. *Urol Clin North Am*. 1992;19(1):149-162.
2. Shergill IS, Arya M, Patel HR, Gill I, eds. *Urological Emergencies in Hospital Medicine*. London, UK: Quaybooks; 2007.
3. Moses AE. Necrotizing fasciitis: flesh-eating microbes. *Isr J Med Sci*. 1996;32(9):781-784.
4. Ayumba BR, Magoha GA. Epidemiological aspects of Fournier's gangrene at Kenyatta National Hospital, Nairobi. *East Afr Med J*. 1998;75(10):586-589.

doi:10.3834/uij.1944-5784.2010.10.12
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5. Murphy M, Buckley M, Corr J, Vinayagamoorthy S, Grainger R, Mulcahy FM. Fournier's gangrene of the scrotum in a patient with AIDS. *Genitourinary Med*. 1991;67(4):339-341.

6. Merino E, Boix V, Portilla J, Reus S, Priego M. Fournier's gangrene in HIV-infected patients. *Eur J Clin Microbiol Infect Dis*. 2001;20(12):910-913.

7. Licheri S, Erdas E, Pisano G, et al. Fournier's gangrene in an HIV-positive patient. Therapeutic options [in Italian]. *Chir Ital.* 2008;60(4):607-615.

8. Elem B, Ranjan P. Impact of immunodeficiency virus (HIV) on Fournier's Gangrene: observations in Zambia. *Ann R Coll Surg Engl*. 1995;77(4):283-286.

9. Eke N. Fournier's gangrene: a review of 1726 cases. *Br J Surg*. 2000;87(6)718-728.

10. Elliott D, Kufera JA, Myers RA. The microbiology of necrotising soft tissue infections. *Am J Surg*. 2000;179:361-366

11. Mergenhagen SE, Thonard JC, Scherp HW. Studies on synergistic infections. I. Experimental infections with anaerobic streptococci. *J Infect Dis*. 1958;103(1):33-44.

12. Benchekroun A, Lachkar A, Bjijou Y, et al. Gangrene of the external genital organs. Apropos of 55 cases [in French]. *J Urol (Paris)*. 1997;103(1-2):27-31.

13. Gonzalez-Ruiz C, Heartfield W, Briggs B, Vukasin P, Beart RW. Anorectal pathology in HIV/AIDS-infected patients has not been impacted by highly active antiretroviral therapy. *Dis Colon Rectum*. 2004;47(9):1483-1486.

14. Hejase MJ, Simonin JE, Bihrle R, Coogan CL. Genital Fournier's gangrene: experience with 38 patients. *Urology*. 1996;47(5):734-739.

15. Kovacs LH, Kloeppel M, Papadopulos NA, Reeker W, Biemer E. Necrotizing fasciitis. *Ann Plast Surg*. 2001;47(6):680-682