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Genital Infection as a First Sign of Acute Myeloid Leukemia

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Key Words
Bacterial infection · Fournier's gangrene · Necrotizing fasciitis · Hematologic malignancy · Acute myeloid leukemia

Abstract
Fournier's gangrene is a life-threatening disorder caused by aerobic and anaerobic bacterial infection. We report a case of genital infection as the initial warning sign of acute myeloid leukemia. We were able to prevent progression to Fournier's gangrene in our patient by immediate intensive therapy with incision, blood transfusions and intravenous administration of antibiotics. This case suggests that hematologists and dermatologists should keep in mind that genital infection can be a first sign of hematologic malignancy.

Introduction

Fournier's gangrene (FG) is a life-threatening disorder caused by synergistic aerobic and anaerobic organisms [1]. The infection of the perineum, scrotum, and/or penis spreads along fascial planes, leading to soft-tissue necrosis [1]. The mortality rate for FG remains high despite antibiotics and aggressive debridement [1]. The initial signs of FG are fever, pain, swelling, and blistering in the genital area [2]. Here, we describe a case of genital infection as the first sign of acute myeloid leukemia (AML).

Case Report

A 51-year-old male presented with fever and a painful, edematous erythema on the scrotum and penis (fig. 1a, b). The eruption had developed after the patient had ridden a bicycle 7 days earlier. Laboratory blood examination results were as follows: white blood cells 7,800/μl with neutrophil 12.0%; lymphocytes 4.7%; monocytes 0.3%; myeloblasts 83.0%; red blood cells 1.97×10^6/μl; platelets 26.4×10^4/μl, and C-reactive protein 4.621 mg/dl. The smear from the bone marrow indicated the presence of a massive myeloblast. The cells expressed CD7, CD11, CD13, CD14, and CD34. An incision was made in the scrotum. *Corynebacterium* spp. was isolated from the genital region. The patient was immediately

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hospitalized and intensively treated with blood transfusions and the antibiotics cefpirome sulfate 4 g and clindamycin 2,400 mg per day for 5 days and cilastatin sodium 2 g and clindamycin 2,400 mg per day for 9 days. The sign of infection diminished two weeks later. Treatment for AML was then initiated using cytarabine, aclarubicin, and granulocyte-colony stimulating factor.

Discussion

FG was mainly caused by trauma and urinary tract infection [3]. In our case, we suspect that the trauma was due to the cycling activity and that the infection was worsened by leukocytopenia due to AML. *Corynebacterium* spp. was cultured from the scrotum. We do not think that *Corynebacterium* spp. was the actual pathologic bacteria because *Corynebacterium* spp. is part of the normal cutaneous flora in the genital area. We believe that our patient showed scrotum infection by unidentified bacteria and that intensive medication prevented progression to ulceration and the more advanced stage of necrosis as a sign of FG.

Few cases of FG have been described as a first sign of hematologic malignancies [4, 5], although cases have been reported as a complication during treatment (**table 1**) [2, 6–11]. Martinelli et al. [2] reported a fetal case having progressive involvement of the abdominal wall resulting in death from leukemia. They indicated that early diagnosis of the disorder and appropriate initiation of an accurate therapy can prevent progression of the acute necrotizing infection [2]. In the reported cases of FG associated with a hematologic malignancy, the edema and swelling were the initial signs [2, 5, 7, 9, 10]. The immediate start of the treatment might have prevented the present patient from being affected by FG.

Hematologists and dermatologists should keep in mind that genital infection and its advanced stage of FG can be an initial sign of hematologic malignancy.

Table 1. Fournier's gangrene or genital infection associated with hematologic malignancies: a summary of the reported cases

Case	Age/sex	Onset of Fournier's gangrene[a]	Hematologic malignancy	Clinical feature of the onset	Clinical feature of the severest situation	Phlogogenous bacteria
Naithani et al., 2008 [6]	17/M	after diagnosis	acute promyelocytic leukemia	painful vesicular lesions in scrotum	ulcers	*Staphylococcus aureus, Escherichia coli*
Mantadakis et al., 2007 [7]	21/M	after diagnosis	acute lymphoblastic leukemia	scrotal edema	a small necrotic scrotal eschar	*Pseudomonas aeruginosa, Staphylococcus epidermidis*
Fukuno et al., 2003 [8]	43/M	after diagnosis	acute promyelocytic leukemia	an ulcer of 0.5 cm in diameter on the left side of the scrotum	swelling that was improved by a surgical incision	no description
Bakshi et al., 2003 [9] 1st case	6/M	after diagnosis	acute myeloid leukemia	edema over the prepuce	a necrotic ulcer	*Pseudomonas aeruginosa*
Bakshi et al., 2003 [9] 2nd case	10/M	after diagnosis	acute lymphoblastic leukemia	pain and swelling in the prepuce	abscess	*Pseudomonas aeruginosa*
Bakshi et al., 2003 [9] 3rd case	9/M	after diagnosis	non-Hodgkin lymphoma	severe pain during micturition, erythema, and tenderness in the penile region	gangrenous changes on the prepuce and glans	no description

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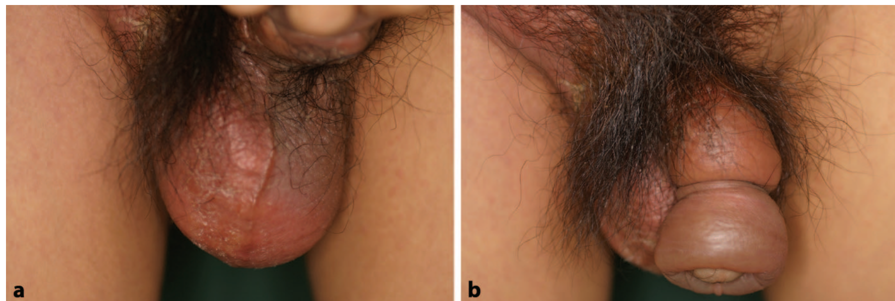
Yoshda et al., 2002 [10]	16/M	after diagnosis	acute myelogenous leukemia	penile swelling with miction pain	gangrene in the regions of the scrotum, penis, thighs, and lower abdomen	*Pseudomonas aeruginosa*
Islamoglu et al., 2001 [5]	33/M	before diagnosis	acute myelomonocytic leukemia	scrotum edema	complete scrotal necrosis, complete penile shaft necrosis, and a left anal ulcer that extended to the left gluteal area	*Bacteroides fragilis*
Martinelli et al., 1998 [2] 1st case	41/M	after diagnosis	acute non-lymphocytic leukemia	genital erythema, pain, swelling and crepitation	blistering and ulceration	*Pseudomonas aeruginosa*
Martinelli et al., 1998 [2] 2nd case	26/F	after diagnosis	acute non-lymphocytic leukemia	redness and swelling of the right labium majorum	ulceration	*Pseudomonas aeruginosa*
Martinelli et al., 1998 [2] 3rd case	26/F	after diagnosis	acute non-lymphocytic leukemia	pain, edema, erythema and swelling of the perineal area	a necrotic ulcer	*Pseudomonas aeruginosa*
Faber et al., 1998 [4]	50/M	before diagnosis	acute myelogenous leukemia	progressive perianal pain	a diffusely infiltrated anal region and bluish scrotum	*Escherichia coli*
Levy et al., 1998 [11]	44/M	after diagnosis	acute promyelocytic leukemia	small indurated lesion of the right scrotum	a painful necrotic area 4×5 cm	*Streptococcus faecalis*, *Staphylococcus* coagulase negative

[a] The onset of Fournier's gangrene or genital infection before or after the diagnosis of a hematologic malignancy.

Fig. 1. a A painful and edematous erythema was present on the scrotum and associated with fever. **b** An edema was observed on the penis.



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