



Post-Hydrocelectomy Fournier's Gangrene in a Patient with Chronic Myeloid Leukemia on Imatinib Chemotherapy: A Case Report

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Abstract

Introduction: Fournier's gangrene is a rare form of rapidly progressing necrotizing fasciitis of the external genitalia and perineal region affecting all ages and gender but mostly older male. Diabetes mellitus, cancer, chemotherapy, HIV infection, inguinal herniorrhaphy and rarely hydrocelectomy are risk factors associated with Fournier's gangrene.

Patient Concerns and Diagnosis: A 56-year-old man on Imatinib for Bcr-Abl positive Chronic Myeloid Leukemia for 15 months underwent uneventful hydrocelectomy 8 days prior to presenting with fever, chills, rigor, scrotal pain and foul-smelling wound discharge. Patient was alert and awake. He had slight tachycardia and tachypnea. The scrotum was enlarged, edematous and tender to touch. Necrotized scrotal tissue with foul smelling discharge was evident. The hydrocelectomy wound was completely dehisced. The white count was normal. Hemoglobin was 9.3gm/dl and platelet count were 69, 000/mm³. Cultures, imaging studies and markers of inflammation were not available. The diagnosis of Fournier's gangrene was made clinically.

Intervention and Outcome: Fluid resuscitation, broad spectrum antibiotics were given. Within 24 hours of presentation, necrotic tissue was vigorously debrided and irrigated. Wound was left open while preserving the testes. Patient recovered well and discharged to follow up with wound closure and further medical care as outpatient.

Conclusion: Fournier's gangrene is a rare and fatal disease that needs emergent care. Clinicians should be aware that FG may be a rare complication of hydrocelectomy like in this case. In mild to moderate cases the outcome may be good, even in resource limited environment, as far as it is recognized early and treated promptly.

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Keywords: Fournier's Gangrene, Hydrocelectomy, Case Report, Chronic Myeloid Leukemia

Abbreviations

CKD: Chronic Kidney Disease

CML: Chronic Myeloid Leukemia

DM: Diabetes Mellitus

FG: Fournier's Gangrene

HBOT: Hyperbaric Oxygen Treatment

VAC: Vacuum Assisted Wound Closure

SGLT-2I: Sodium Glucose Cotransporter Two Inhibitor

Introduction

Fournier's gangrene (FG) is a rapidly progressive type of necrotizing fasciitis of the external genitalia and perineal region associated with high case fatality rate. It took its name from five cases of perineal gangrene described in 1883 by the French dermato-venereologist Jean Alfred Fournier [1-3]. The disease affects all ages and gender but mostly older men [4,5].

Risk factors for FG, among others, include diabetes mellitus, cancer and chemotherapy. Only three clear cases of FG associated with hydrocelectomy have been described in the literature [6-8]. Clinical presentation of FG may include fever, chills, painful scrotal swelling, erythema and foul-smelling discharge. Exam may show crepitation and skin necrosis. When available, laboratory work up and imaging studies may help in diagnosis, mortality risk stratification and decision in surgical approach [2,5,9]. Infection in FG is mostly caused by polymicrobial organisms. Infection with multidrug resistant bacteria has been well documented [10,11]. Treatment includes volume expansion, broad spectrum antibiotics and surgical debridement within 24 hours. Repeat debridement, ancillary surgery such as diversion colostomy and reconstructive surgery may be necessary. Hyperbaric oxygen therapy (HBOT) and vacuum assisted closure (VAC) are useful in selected cases but not universally required [2,3,9]. Outcome from FG may have been improving due to better antimicrobial agents, guideline-based care and improved surgical techniques. Lower mortality rate from FG has been reported from resource limited setting such as Sub-Saharan Africa

presumably due to young patient age and infection with less resistant organisms [14,12,13]. To our knowledge, post-hydrocelectomy FG has not been described from Ethiopia were the index case is reported and from Sub-Saharan Africa at large. In this article, we present and briefly discuss the case of a 56-year-old patient with chronic myeloid leukemia (CML) on treatment in remission phase who developed Fournier's gangrene after hydrocelectomy. Clinicians should be aware that FG may rarely occur as infectious complication of hydrocelectomy but outcome is generally good as far as detected and managed early.

Case Repots

A 56-year-old man with the past medical history of Bcr-Abl positive chronic myeloid leukemia on Imatinib for 15 months presented to hospital with uncomfortable bilateral scrotal swellings. After routine examination he was diagnosed with bilateral hydrocele. Hydrocelectomy was successfully done electively and drainage was kept in situ for 48 hours which was removed before discharge from hospital. No prophylactic antibiotic was given. 8 days later, he presented with subjective complaints of fever, chills, rigor, scrotal pain, wound discharge and wound dehiscence. He was alert awake with stable vital signs except mild tachycardia and tachypnea. The scrotum was swollen, edematous and tender to touch. Necrotized tissue with foul smelling discharge was evident. The rest of physical examination findings were unremarkable. Blood test showed a white count of 3800/mm³ with absolute neutrophil count of 1800/mm³. Hemoglobin was 9.3gm/dl and platelet count were 69, 000/mm³. Chemistry, inflammatory markers, lactic acid level, cultures and imaging studies we're not available due to limited resource. Fournier's gangrene was diagnosed clinically. Intravenous fluid resuscitation, broad spectrum antibiotics were started. He had scrotal wound debridement and irrigation within 24 hours of presentation. He recovered well and discharged home to undergo delayed wound closure. Image of healing debrided scrotum is shown in Figure 1 below.



Figure 1. Post-debridement image of Fournier's gangrene of scrotum that developed after hydrocelectomy.

Discussion

Fournier's gangrene infection is thought to begin in the subcutaneous tissue. Infection is mostly polymicrobial although single microbial and multiple drug resistant bacterial infection can occur often [10,11]. The bacterial toxins produced, end arterial and venous thrombosis lead to tissue destruction and necrosis more than it appears on the skin surface. In severe cases, disease can spread along loosely interconnected anatomical fascial planes of the perineum, thighs and abdomen [2,3,14]. Like in our patient, FG typically affects men older than 50 years. Predisposing factors, among others, may include diabetes mellitus, hypertension, renal failure, cancer and chemotherapy mostly via impaired micro-circulation or depressed immunity [2,3,5,9]. Our patient had CML diagnosed 15 months prior. He was on Imatinib, a tyrosine-kinase inhibitor that has been linked to cytopenias, occasional infection and delayed wound healing but not with FG. Although our patient had bicytopenia, his white blood cell and absolute neutrophil counts were in normal range. Both Imatinib and CML could be implicated as predisposing factor for FG in our patient but no similar case report exists for comparison. In the English literature, at the time of this case report, there were only four case reports of FG in post-hydrocelectomy patients (6-8) (table 1).

Table 1. Cases of post-hydrocelectomy Fournier's Gangrene reported in the literature

Case Author and reference	Patient age in years	Comorbidity	Days post hydrocelectomy	Outcome
Al-Ali BM ⁶	78	None	15	Alive
Sahu M et al ⁷	79	Hypertension, CKD	09	Alive
V, K. G ¹⁵	40	Type I DM	30	Alive
Vargo E et al ⁸	64	Type 2 DM, on SGLT-2 I	18	Alive
Index case	56	CML, on Imatinib	08	Alive

CKD chronic kidney disease, DM diabetes mellitus, SGLT-2 I sodium glucose co-transporter 2 inhibitor.

In one case, FG occurred 30 days after healed hydrocelectomy in type I diabetic patient [15]. Hydrocelectomy may not necessarily be a risk factor for FG in this patient. In the remaining three cases, one patient had no associated medical comorbidities. Of the other two patients, one patient had hypertension and CKD as associated comorbidity with FG, whereas the second patient had diabetes treated with Sodium glucose Cotransporter-2 (SGLT-2) inhibitor as associated comorbid factor with FG. SGLT-2 inhibitor use has been linked to FG in case series [16]. FG is a clinical diagnosis and in resource limited setting, like in our case, spectra of lab, histological examination and imaging studies may not be available making management in severe cases challenging. Treatment generally includes fluid resuscitation, broad spectrum antibiotics and surgical debridement within 24 hours. Repeat debridement, ancillary and reconstructive surgeries may be necessary. Although not universally available or required, HBOT and VAC are useful in selected cases [2,9]. Our patient received volume resuscitation, broad spectrum antibiotics and timely wound debridement. He was scheduled to undergo delayed wound closure. Mortality rate in FG ranges from 20% to 40% but in more recent data, including Sub-Saharan Africa, the figure may be lower than that [4,12,13]. In all reported cases of post-hydrocelectomy FG, including our patient, outcome was excellent perhaps due to early detection as a result of post-hydrocelectomy follow up [6,8]. In conclusion, FG is a rare disease with spectrum of presentation ranging from clinically mild to moderate cases like in our case to a more extensive and fatal form requiring multidisciplinary care. Clinicians should be aware that Fournier's gangrene may rarely complicate hydrocelectomy, especially in patients with medical comorbidities, like the index case but the outcome appears good, even in resource limited environment, as far as it is recognized early and treated promptly.

Strength and Limitation

This case report adds information to the available literature in that Fournier's gangrene could rarely complicate hydrocelectomy especially in patients with medical comorbidities. With post-procedure follow-up and early recognition, good outcome could be expected. Since this is a single case report inference or cause effect association of CML, hydrocelectomy alone or in combination with FG could not be made

We also acknowledge that biopsy results and other more extensive tests were not possible due to resource limitation.

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