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CASE REPORT

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Fournier's gangrene after missed acute perforated appendicitis: A case report

Ismail Elahabadi[1] | **Gholamreza Bazmandegan**[2,3] | **Hossein Salehi**[1] | **Amin Jafari**[1] | **Jafar Ahmadi**[4] | **Zahra Kamaib**[2,3]

[1]Department of Surgery, Ali-Ibn Abi-Talib Hospital, School of Medicine, Rafsanjan University of Medical Sciences, Rafsanjan, Iran

[2]Clinical Research Development Unit, Ali-Ibn Abi-Talib Hospital, Rafsanjan University of Medical Sciences, Rafsanjan, Iran

[3]Department of Family Medicine, Ali-Ibn Abi-Talib Hospital, School of Medicine, Rafsanjan University of Medical Sciences, Rafsanjan, Iran

[4]Department of Radiology, Ali-Ibn Abi-Talib Hospital, School of Medicine, Rafsanjan University of Medical Sciences, Rafsanjan, Iran

Correspondence
Zahra Kamiab, Clinical Research Development Unit, Ali-Ibn Abi-Talib Hospital, Rafsanjan University of Medical Sciences, Rafsanjan, Iran.
Email: dr.kamiab89@gmail.com

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Abstract

Fournier's gangrene (FG) is a rare progressive necrotizing fasciitis (NF) with high mortality rate. This case report describes a young patient with FG with no known history of disease or invasive therapeutic interventions.

KEYWORDS

Fournier's gangrene, necrotizing fasciitis, retroperitoneal appendicitis

1 | BACKGROUND

Necrotizing fasciitis (NF) following missed acute perforated appendicitis is a potentially life-threatening condition. In patients with retroperitoneal appendicitis, this condition can lead to Fournier's gangrene (FG). This study aimed to report a young patient with FG with no known history of disease or invasive therapeutic interventions. Fournier's gangrene is a rare but rapidly progressive NF of the perineum with high mortality rate.[1] It is in fact an acute urological emergency, and its mortality rate is between 15% and 50%.[2] It is characterized by subcutaneous vascular thrombosis, the progression of the gangrene, and subsequent infection process. Predictive factors for FG include old age, diabetes mellitus, obesity, cardiovascular disorders, chronic alcoholism, long-term corticosteroid therapy, malignancy, and those with weakened immune systems (such as HIV infection). Urinary tract obstruction and trauma have also been associated with the disease.[3] In general, organisms of the urinary tract expand along the fascia to involve the penis and scrotum, the gluteal region, and the anterior abdominal wall.[4]

The disease has a wide range of clinical manifestations; it can have a slow and progressive onset or a sudden onset with rapid progression. Local symptoms include swelling and redness associated with pain and tenderness, and possibly crepitus. Skin inflammation gradually worsens and progresses to spotty necrosis and gradually widespread subcutaneous necrosis.[5] Differential diagnosis of FG includes scrotal and perineal disorders, as well as intra-abdominal disorders, such as cellulitis, scrotal abscess, strangulated hernia, pyoderma gangrenosum, allergic

vasculitis, vascular occlusion syndrome, and Warfarin necrosis.[6]

A delayed diagnosis of FG, due to the rapid progression of the disease, leads to sepsis, multiple organ failure, and disseminated intravascular coagulation, and ultimately to very high mortality. Immediate diagnosis and appropriate treatment consist of broad-spectrum antibiotics, extensive surgical debridement, and hemodynamic support to reduce complications and control infection.[7]

Even a few hours of delay in surgical debridement in the event of FG dramatically increases the risk of death. Therefore, as soon as the diagnosis is made, an immediate surgery should be performed.

Acute appendicitis has been identified as another cause of FG, and it is a rare pathology. Despite the limited number of reports in this regard,[8,9] the aim of this study was to report a patient with NF and FG with no known history of disease or invasive therapeutic interventions.

2 | CASE REPORT

A 25-year-old man was referred to the emergency department 15 days before hospitalization due to epigastric pain, fever, chills, and oral intolerance. After a while, the patient's symptoms decreased and he was discharged. But after 5 days, due to a pain in the right flank area with a diffusion to testis with a pain in the hypogastric region, dysuria, scrotal swelling, and the discoloration of the urine, he was referred to an urologist and was referred to the emergency department again by the urologist. At the time of admission (September 2018), the patient had fever, chills, dyspnea, chest pain, and myalgia. He did not report a history of specific illness in himself or his family, drug use, allergies, smoking, and addiction.

The results of examination were as follows: PR: 135, RR: 28, BP: 70/50, T: 37.5°C.

The examinations of heart and lung were normal. On examination, the abdominal wall in the right flank area had an erythema and was soft to the touch. A tenderness was in the hypogastric region and in the lower right abdominal area, he had the ecchymosis of the left scrotum without crepitation. Also, there was an anaerobic odor (Figure 1). The results of laboratory tests are reported in Table 1.

After the initial examination, due to tachycardia and chest pain, a cardiac consultation to rule out PTE was performed and the patient underwent CT angiography, after which the PTE was rejected for the patient. In cardiac consultation, due to evidence of myocarditis and EF: 25%–30% on echocardiography, it was recommended to start dopamine drip, Lasix, digoxin, and ICU care.

In an advice on the infection, urologic consultation for FG was recommended. In the urology consultation, given

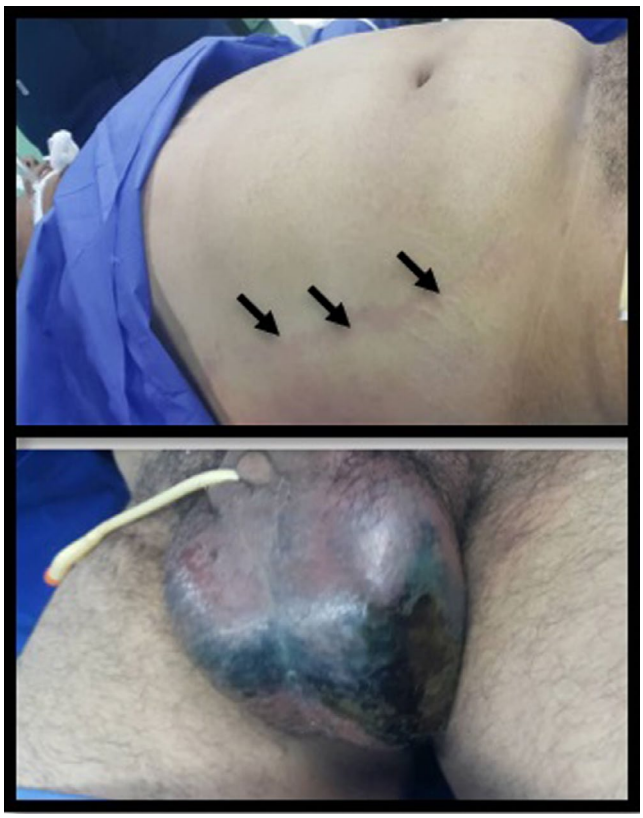

FIGURE 1 Erythema of right flank area and ecchymosis of the left scrotum

the ecchymosis of the left scrotum and the anaerobic odor and bilateral scrotal swelling, the onset of FG was raised. After a genital wash and the administration of ciprofloxacin and metronidazole, an emergency surgery was recommended for FG, considering the cardiac service and ultrasound for a diagnosis in the soft tissue, the scrotum, and inguinal region. Before the surgery, ultrasound and CT of the abdomen and pelvis were performed without contrast. In an early ultrasound, an evidence of inflammation was seen in the right flank and no evidence of appendicitis was found. For this reason, a CT scan was performed, which showed myositis and fasciitis in the soft tissue of the right flank and inguinal region as well as a collection in the lower right abdominal area, and according to the radiologist, appendicitis was reported. The CT scan also showed multiple small collections in the lower right abdominal area and air bubbles in the anterior part of the psoas muscle, and myositis and fasciitis in the anterior outer wall of the abdomen, and necrosis and inflammation in the right inguinal and abdominal wall. Due to the fulminant course of NF of the right flank and gangrene of Fournier in the early hours of admission to the emergency department, diagnostic laparoscopy was not possible. After CT scan and evidence of complicated appendicitis and necrotic tissue in the abdominal wall and scrotum and septic shock created for the patient, emergency

TABLE 1 Laboratory tests of the patient at the beginning of the visit

		Laboratory test
WBC ($\times 10^6$/L)		20,000
HB (g/L)		14
MCV (fl)		85
PLT ($\times 10^6$/L)		70,000
CRP (mg/L)		42
ESR (mm/h)		32
CPK (U/L)		525
Urea (mg/dl)		29
Creatinine (mg/dl)		0.8
VBG	PH	7.489
	PCo2(mmHg)	22.1
	Po2 (mmHg)	61.8
	HCo3 (mEq/L)	16.7
	O2 sat (%)	93.4

surgical consultation was requested. According to the patient's emergency condition, laparotomy and debridement of necrotic tissue were performed.

The patient underwent an exploratory laparotomy, the debridement of the necrotic tissue and the scrotal area, as well as the discharge of intra-abdominal abscesses.

After leaving the operating room due to septic shock, the patient was transferred to the ICU and was placed under the joint care of the surgical and infection control services to continue treatment. Due to septic shock, dopamine drip was prescribed for the patient. After 3 days, the patient's pressure drop recovered and dopamine drip was gradually discontinued. On day 7, due to a pleural effusion and the ARDS evidence, the patient underwent a diagnostic pleural tap, a pleural fluid culture, and a thorax CT scan, and no evidence of empyema was found. A chest tube was inserted for the patient, and finally, with regard to the pleural fluid culture, *E. coli* was raised.

On day 10, the patient underwent re-echo due to continued tachycardia, and due to EF: 45%, he was advised to take metoral for treatment. He was extubated on day 12. Five days after chest tube insertion, chest tube was removed due to reduced secretions. After 1 month, the patient underwent skin grafting due to the improvement in his general condition and the cut tissue. However, due to continued tachycardia, he underwent echo before reoperation, and according to EF: 30%–35%, carvedilol, aldactone, and aspirin were administered (Figures 2 and 3).

Finally, after the improvement in his general condition, he was transported to the operating room for surgery and underwent split-thickness skin grafting in the right flank

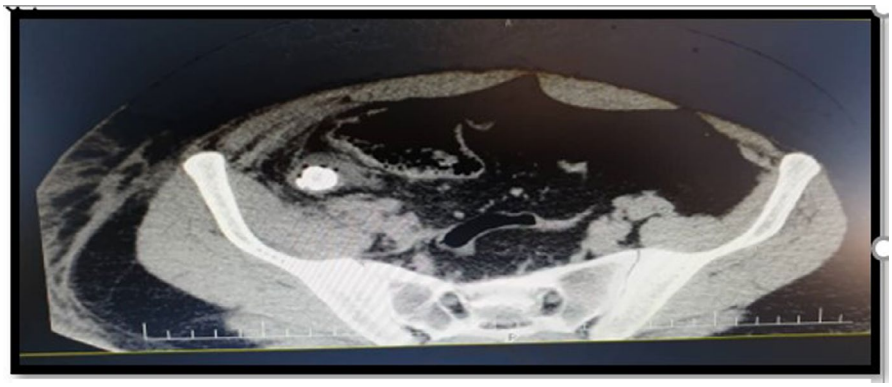

FIGURE 2 Gas accumulation and soft tissue in the patient's CT scan with a diagnosis of FG and NF of the abdominal wall following perforated appendicitis (Flash location: edema and inflammation of soft tissue and gas in the abdominal wall)

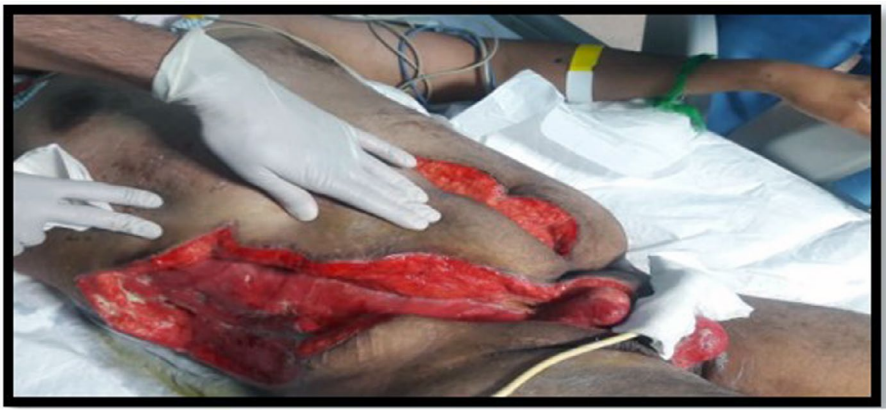

FIGURE 3 Debridement of infected tissues and patient's wound conditions after the surgery for NF of the abdominal wall

area and genital area. After the graft recovery, the patient gradually had RBR and was discharged in good general condition after 30 days.

The study was approved by the Ethics Committee under the code: (IR.RUMS.REC.1399.100), in accordance with Helsinki Criteria.

3 | DISCUSSION

Fournier's gangrene or NF of the perineal region is a challenging situation in surgery. Tissue ischemia is a major pathogenic factor, and usually progresses to gangrene with the rapid progression of myonecrosis. The culture of necrotic tissues shows aerobic and anaerobic bacteria, including Coliforms, Klebsiella, Streptococcus, Staphylococcus, Clostridia, Bacteroides, and Corynebacterium.[10] In this case, the tissue culture sample showed *E. coli*.

The synergistic activity of aerobes and anaerobes results in the production of various exotoxins and enzymes, such as collagenase, heparinize, hyaluronidase, streptokinase, and streptodornase, leading to tissue destruction and the spread of infection. The platelet aggregation and complement fixation induced by the aerobes and the heparinize and collagenase produced by the anaerobes lead to microvascular thrombosis and dermal necrosis.[11] Due to the gradual process of getting the tissues affected by the bacteria, the onset of the disease is dramatically deceptive. The subcutaneous fat and fascia are initially affected, and no evidence of disease is apparent on the surface of the skin, and only a localized erythema may be seen, which is often confused with cellulite at first. In this case, only local erythema was also seen on the surface of the abdomen, but in the scrotum, ecchymosis was significant.

As the disease progresses, the skin turns gray and black and looks burnt. In this situation, the patient's condition deteriorates rapidly, and any delay in the diagnosis, the initiation of drug treatment, and the surgical debridement of necrotic tissue can lead to death. The mortality rate in patients treated within 24 h was 12.5%, but it was 72.7% in those whose surgeries were delayed for 4 days.[12] Although the overall mortality rate has not changed since the beginning of antibiotics use, and their effectiveness has not been established, it is recommended that broad-spectrum antibiotics be started before the sensitivity results. It is important to monitor the patient's condition for hypovolemia and shock. In case of occurring extensive edema due to NF, as well as open skin wounds after surgery, the patient is susceptible to dehydration and protein loss. Aggressive supportive treatment is also important, including respiratory support, central cardiovascular monitoring, hemodialysis, and oral or intravenous nutrition. The most common causes of death from NF include sepsis, disseminated intravascular coagulation, respiratory failure, kidney failure, and multi-organ failure.[13] It is, therefore, important that the physician does not delay the diagnosis of the disease in an attempt to prevent the consequences of the disease, such as extensive NF or its possible fatal consequences. The patient with clinical suspicion of NF underwent rapid diagnostic and therapeutic workup, and the above-mentioned complications were not observed in him. Acute appendicitis is known to be another cause of FG, which is mainly secondary to viscous rupture in the posterior or posterior abdominal spaces with the spread of infection to the perineum and scrotum.[14]

The most important predictors and causes of clinical suspicion of the disease are diabetes and alcoholism. Other important contributing factors to diagnosis include internal catheters, local trauma, surgery, malignancy, steroid use, chemotherapy, radiotherapy, prolonged hospitalization, and human immunodeficiency virus. The important point in the patient was the history of his diseases, which was negative, and the only history of the patient was an epigastric pain that continued with the right flank pain. On ultrasound, there was an evidence of perforated appendicitis. Previous studies have described NF as a potentially life-threatening disease in patients with missed perforated appendicitis. This complication rarely occurs.[15] Another important point is that the retroperitoneal appendix, located in the right anterior pararenal space, is associated with the pelvic space,[16] and its infection and inflammation can transmit to the prevesical space, thereby causing ischiorectal abscesses. A rare case of this spread of infection is FG[17] reported in the patient.

This case highlights the importance of clinical suspicion for any patient who does not even have the major risk factors for FG and does not have a history of a serious illness. Given FG is a rare but potentially life-threatening complication of missed perforated appendicitis, its diagnosis in young patients with early symptoms of NF and FG and a history of a gastrointestinal pain similar to an acute appendicitis pain and its subsequent timely management for infection control, necessary surgery, and cardiovascular support are vital.

It should be noted that if patients with suspected appendicitis are evaluated on their first visit to the emergency room based on guidelines for diagnosis and evaluation of abdominal pain, they will never miss and will not be diagnosed late. However, due to the fact that the appendix was retrocecal and did not meet the Alvarado's criteria and was admitted to the emergency room with shock sepsis and FG, emergency laparotomy was needed.[18]

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

I E made substantial contributions to acquisition of data and revised the manuscript critically for important intellectual content. Z K conceptualized and designed the study, made substantial contributions to conception and acquisition of data, reviewed the manuscript critically for important intellectual content, approved the final version, and collaborated on article revision H S, A J, and J A made substantial contributions to acquisition of data G B reviewed the manuscript critically for important intellectual content, approved the final version, and collaborated on article revision.

CONSENT

The patient provided written ethics consent for publication.

DATA AVAILABILITY STATEMENT

Research data are not shared.

ORCID

Zahra Kamaib https://orcid.org/0000-0001-6670-1828

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