Case Report

Diagnosis and treatment of Fournier's Gangrene: Two cases and literature review

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ABSTRACT

Fournier's gangrene (FG) is a rapid progressive disease with high mortality and is caused by polymicrobial infection. FG usually begins with infection and affects fascias in (the perianal and) perineal regions as well as the abdominal wall and other organs. Although this disease has been recognized for many years, there are only a few cases reported in the world and few from China. Here, we report our success in the diagnosis and treatment of two cases of FG, one of which had severe necrotic sing fasciitis spreading to the abdominal wall, perianal, and perineal regions. We have discussed the pathogenesis, diagnosis, and treatment of FG.

KEY WORDS: Fournier's gangrene, Perianal abscess, Diagnosis; Treatment.

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INTRODUCTION

Fournier's gangrene (FG) is a fulminant necrotizing fasciitis of the genital, perianal, and perineal regions and characterized by rapid progression and high mortality.^{1,2} FG is usually caused by severe bacterial infection in subcutaneous tissue and superficial fascia, especially in the scrotum and penis, which can result in obliterative endarteritis and thrombosis of small arteries.^{1,2} Other factors, including older age, diabetes mellitus, malignancies, immunosuppression, and chronic alcohol abuse, also contribute to the development of FG.^{1,3}

Despite the increasing improvement in diagnosis and therapy of FG, the mortality of FG remains high. In the past 40 years, there are only a few cases with FG reported in the world, and few from China.

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Importantly, there is no standardized protocol for the management of FG patients. Recently, we have successfully diagnosed and treated two patients with FG. Here, we report these two cases and briefly review the FG-related literatures.

CASE REPORT

Case-1: The patient is a 45-year-old man. He complained of anal pain for a couple of days and was diagnosed with perianal abscess, followed by incision and drainage of perianal abscess at a suburban clinic. Two days later, he was admitted to our hospital because of the presence of continual fever and perineal and scrotal ulcers. He denied any previous history of chronic diseases, such as diabetes and diseases leading to immunodeficiency, and he did not take any immunosuppressive drugs during the past year. Physical examination revealed a body temperature of 38.5°C and blood pressure of 120/90 mmHg, without any obvious abnormality of the heart and lungs. Rectal examination of the patient at the Bozeman's position found two openings at the 3 and 9 o'clock of the anus, from which yellow fetor pus was noticed (Fig-1).

The surrounding skin and deep fasciae were dark and covered with much purulent and necrotic debris. His scrotal skin had an ulcerative defect of $6 \times 5 \times 5$ (cm) with foul purulence, and the internal fascia was also covered with fetor pus and necrotic debris. His testes were only partially covered by the scrotal skin, but with superficial yellow purulence. The laboratory examination indicated a white blood cell count of 16.43×10^9 /L, 77% neutrophils. The concentration of serum total protein was 62.2 g/L; albumin, 31.3 g/L; acetylcholine esterase, 2645 U/L; and Na+: 131.70 mmol/L. The patient was diagnosed with FG on the basis of his perineal and scrotal ne-crotizing fasciitis and perianal abscess. Accordingly, the patient was treated intravenously with 3.0 g of Cefoperazone and 0.2 g of Metronidazole, and subjected to surgical debridement and drainage in the perineal and perianal regions by a Penrose drain.

During the surgery, we found that the two abscesses were deep toward the ischiorectal space, accompanied by many embolisms in small subcutaneous vessels, severe fascial swelling, and extensive necrosis, but did not spread to the local muscles. We removed the necrotizing skin and fascial tissues from the lesions up to the healthy tissue on the inner wall. Furthermore, his wound were washed sequentially with 5% hydrogen peroxide, 0.2% Metronidazole and Gentamicin, and saline repeatedly, and then covered with iodoform gauze. After confirmation of Escherichia coli infection from bacterial culture of the purulence, the patient was intravenously treated with 3.0 g of Cefoperazone twice per day and 0.2 g of Metronidazole once per day for seven consecutive days, according to the results of the antibiotic sensitivity of Escherichia coli.

Simultaneously, his wounds were continually cleaned and washed daily. One week after surgery, his laboratory examination returned to normal ranges and his infection subsided. Subsequently, his wounds grew with fresh granulation and gradually healed. He was discharged from hospital 38 days after admission with complete recovery.

Case-2: The patient was a 19-year-old man and complained of consistent abdominal pain and distension with fever. He first had abdominal and perineal pain for 10 days, and was treated with the perianal debridement and drainage for his perianal abscess at a local clinic. However, his symptoms did not improve and he was transferred to our hospital. He also denied any chronic history of diabetes and long-term use of immunosuppressive drugs. Physical examinations revealed that his vital signs were normal, body temperature: 37.6 °C, and blood pressure: 118/ 70 mmHg. However, his bilateral buttocks were asymmetrical, and his right buttock was red swelling, spreading to the perinaeum and scrotum in approximately 10 cm×7 cm×8 cm. Foul purulence was drained from his right scrotum, and the hypogastric

wall and lower abdomen was red swelling, with tenderness, rebound tenderness, and muscular tension without clear margin. Laboratory examinations indicated that he had a white blood cell count of 18.24×10⁹ /L, and 79.4% neutrophils.

The concentration of his serum total protein was 31.0 g/L; albumin, 13.8 g/L; acetylcholine esterase, 1154 U/L; and Na+: 123.70 mmol/L. A computed tomography of the abdomen and pelvis indicated extensive emphysema around the scrotum (Fig-2A), fluid in the ischiorectal space, around the bladder and in the abdominal wall with emphysema (Fig-2B and C). Accordingly, the patient was diagnosed with FG (acute necrotizing fasciitis and pararectal, scrotal, and abdominal abscesses). The patient was subjected to debridement and drainage of the perianal position, abdominal wall, and scrotum and treated with antibiotics of 3.0 g of Cefoperazone and 0.2 g of Metronidazole. The patient was laid in the lithotomy position, and the anal 3 and 7 o'clock positions of the perineal region were cut open under local anesthesia. About 1000 ml of the foul and brown purulence was drained out from the incision. The abscess cavity reached the bilateral ischial tuberositics, posterior to the sacral bone, and anterior to the articulation of pubis, respectively. To drain out the purulence in the abdominal abscess, an incision was performed along the hypogastric median. Extensive subcutaneous necrotic tissues were removed and another 1000 ml of fetor purulence was drained out. The abscess cavity in abdominal wall reached to the intersection of bilateral midaxillary lines and costal archs, and extended to the ischiorectal space through the posterior space of the pubic symphysis.

There were many embolisms of small subcutaneous vessels around the perianal and abdominal incisions, and extensive necrotic fascias, which were separated from muscular tissues. Accordingly, we completely removed the necrotic skin, subcutaneous



Fig-1: The necrotic scrotal skin, perianal abscess, and the affected testes.

tissues, and deep fascia up to the healthy muscle tissues. Furthermore, we repeatedly washed the abscess cavity with 5% hydrogen peroxide, saline, and 0.2% of Metronidazole daily, followed by inserting several drainage tubes into the abscess cavity. After confirmed infection with Klebsiella pneumoniae by culture study, the patient was treated intravenously with 3.0 g of Cefoperazone twice per day and 0.2 g of Metronidazole once per day for 10 consecutive days, according to the antibiotic sensitivity profile, supplemented with supportive therapy. Subsequently, the new necrotic tissues in his wound were immediately removed and washed daily. Five days after surgery, fresh granulation appeared in the cavity, and two days later, his trauma was significantly reduced, accompanied by normal laboratory examinations. Eleven days after surgery, the patient was discharged and returned to his hometown for continual supportive treatment. One month later, he was completely cured.

DISCUSSION

FG (FG) is a rapid, progressive, and fulminant form of necrotizing fasciitis in the genital, perianal, and perineal regions, and is commonly caused by polymicrobial infections. In some cases, necrotizing fasciitis in FG patients can extend towards the abdominal wall between the fascial planes. Previous studies have shown that the development of FG usually starts with anorectal and urogenital trauma and infections, particularly in aged individuals with diabetes, immunodeficiency, malignant tumor, obesity, alcoholic addiction, organ transplantation, chronic hepatic disease, malnutrition, HIV infection, and neutropenia,^{1,4}

The FG-related infection is commonly caused by multiple types of bacteria, predominantly including *Escherichia coli*, *Pseudomonas aeruginosa*, *Klebsiella species*, *Streptococcus species*, *anaerobes Bacteroides*

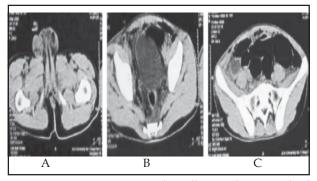


Fig- 2: CT images. A: Scrotal swelling and gas; B: Fluid and gas in ischial-rectal space, paravesical space, and underneath abdominal wall; C: Fluid and gas underneath abdominal wall.

fragilis, Clostridium perfringens, and *Candida glabrata,* which usually inhabit the lower gastrointestinal tract or the perineum.³ Pathophysiological researches have suggested that invading bacteria stimulate inflammation, and recruit inflammatory neutrophils and macrophages that produce inflammatory cytokines, lysosome, and radical oxygen species (ROS), leading to resultant obliterative endarteritis, thrombosis of the cutaneous and subcutaneous vessels, and tissue necrosis.⁵ The infection and related inflammation can rapidly spread along the fascial planes, because of the unique characteristic of fascial structures in genital, perianal, and perineal regions as well as the abdominal wall.

Because of the rapid progression and high mortality, early diagnosis is critical for the control of FG. Clinically, patients usually display erythema, edema extending beyond erythema, severe pain, and livid discoloration of the skin in the local lesions of the perianal, perineal, and genitalia scrotal region, accompanied by systemic toxic symptoms, such as high fever, increased white blood cell counts, and high frequency of neutrophils. Careful examination should find the abscess formed, possibly with fetor purulence. However, early symptoms may be atypical and difficult to be accurately diagnosed. Our two cases had no diabetes and other risky susceptible conditions for the development of FG. Both cases began to display perianal abscess, and one patient developed necrotic fasciitis with a huge abscess that had spread to the abdominal wall. They were not diagnosed with FG until they were transferred into our hospital, possibly due to the lack of disease recognition and the physician's inexperience. However, the failure of early diagnosis usually leads to rapid spreading of FG to deep fascial tissues.

Evidently, we found that the perianal abscess in the second patient had spread to the abdominal wall up to the epigastric region. Indeed, extensive necrotic fascial tissues and thrombosis of the microvasculature were found in the patient, accompanied by huge amounts of fetor purulence. Therefore, we should pay special attention to the patients with symptoms and physical signs of perianal and perineal abscess, and consider the potential of FG when we make differential diagnosis. Furthermore, increased white blood cell counts (>15.4×10⁹/L), elevated levels of plasmic creatine phosphokinase, decreased concentration of sodium (<135 mmol/L), uncontrolled high fever, exacerbated systemic conditions are the signs of potential FG⁶⁻⁷ and the laboratory risk indicators for necrotizing fasciitis score may be used as references although their value remains debateable.8 In addition, CT and MRI examinations are valuable for diagnosis and determination of FG severity.⁹⁻¹⁰

Importantly, intraoperative and postoperative pathological examinations of the skin, subcutaneous fat, and fascial tissues are of significance in the diagnosis of necrotizing fasciitis. Indeed, we examined the second patient by CT and observed extensive emphysema around the scrotum, fluid in the ischiorectal space, around the bladder, and in the abdominal wall with emphysema. Early diagnosis of FG may be helped by the "finger test".⁵ These, together with the patient's history of chronic diseases and long-term medication, will be crucial for early and accurate diagnosis of FG.

Immediate and aggressive surgical treatments, such as surgical debridement and drainage, are critical for the management of FG. We conclude that when FG is clinically indicated, repeated debridement should be performed. We cleaned the wound daily for the first couple of days, because the extensive abscess had spread up to the epigastric region of the abdomen. Furthermore, we washed the wounds sequentially with hydrogen peroxide, antibiotic solutions, and saline repeatedly, and inserted several drainage tubes into the abscess cavity. These aggressive surgical debridement and drainage not only immediately removed the necrotic tissues and inhibited bacterial growth, but also prevented the absorption of toxic factors produced by bacteria and necrotic tissues, facilitating the recovery of patients. Our findings support the notion that repeated aggressive debridement can reduce the mortality of FG patients.11

In addition, we believe that treatment of the patients with optimal antibiotics is important to control the progression of FG and to promote rapid recovery. We treated the patients intravenously with broad-spectrum antibiotics, such as third-generation 3.0 g of Cefoperazone twice per day and 0.2 g of Metronidazole, before receiving microbiological conformation and drug sensitivity and changed to pathogen-sensitive Cefoperazone and Metronidazole after microbiological tests. This intravenous combined antibiotic therapy should effectively prevent the infection from spreading to other organs, reducing the development of FG-related fatal complications. Moreover, the maintenance of water-electrolyte balance and acid-base equilibrium, together with nutritional therapy, benefits the rapid recovery of our patients. We used these therapeutic strategies and achieved a successful cure of FG within a short period. Recently, other therapeutic strategies, such as the hyperbaric oxygen therapy and drotrecogin⁹

treatment, have been suggested for the treatment of FG because of their anti-inflammation and antithrombosis activities.¹¹⁻¹² Their therapeutic efficacy remains to be further determined. Finally, Stage second hermatoplasty and skin transplantation may help in the recovery of patients, particularly for those with extensive cutaneous deficiency.

In summary, perianal or perineal abscess is a common infectious disease, which can be complicated by acute necrotizing fasciitis in the perineal region, scrotum, and abdominal wall, leading to the development of FG, a rapid progressive disease with high mortality. We recommend the early diagnosis of FG by the combination of clinical manifestation, Lab examination, "finger test", CT, and MRI. We experienced that immediate aggressive surgical debridement and drainage from the trauma, together with the broad-spectrum or pathogen-sensitive antibiotics, water-electrolyte balance, acid-base equilibrium, and nutritional support, were critical for the treatment of the disease.

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