



ORIGINAL ARTICLE

Outcomes in patients with Fournier's gangrene originating from the anorectal region with a particular focus on those without perineal involvement

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Abstract

Background and aim: Fournier's gangrene (FG) is a fulminant infection in the external genital region and perineum. The present study explored the clinical features of FG originating from the anorectal region, from primary conditions such as anal fistulas and abscesses.

Methods: A retrospective analysis was performed in order to identify the factors associated with clinical outcomes in FG patients derived from two hospitals—the Sixth Affiliated Hospital of Sun Yat-sen University and People's Hospital Affiliated to Fujian University of Traditional Chinese—over the period from May 2013 to April 2017.

Results: Sixty FG patients were included in this study. The common causative microorganisms cultured were *Escherichia coli* species. Genital and perirectal regional involvement was evident in 52 and 59 cases, respectively, although the perineum was unaffected in 7 cases (12%), as confirmed by imaging examination and surgical exploration. Management with early radical debridement and broad-spectrum antibiotic therapy is effective with an acceptably sepsis mortality (1.7%). Ten patients underwent protective colostomy. No patient underwent an orchidectomy and required urinary diversion.

Conclusions: FG originating from the anorectal region can be rapidly progressive and life-threatening. Infection can spread superiorly to the genital region without the involvement in perineal tissue. An aggressive surgical debridement of non-viable tissue is essential for satisfactory outcomes and a protective colostomy is not mandatory.

Key words: Fournier's gangrene; perianal abscess; anal fistula; protective colostomy

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Introduction

Fournier's gangrene (FG), first described by Fournier in 1883, is a fulminant infection of the external genital region and perineum resultant from mixed aerobic and anaerobic bacteria [1]. It is an uncommon condition that can affect patients of any age and can rapidly progress to a life-threatening stage. The incidence of FG remains low, with reported rates ranging from 1/7500 to 1/2000 in hospitalized patients and with a reported mortality rate of 16% [2]. Diabetes mellitus and immunosuppression are two major risk factors for FG [3], with anorectal and urogenital infections and traumas being amongst the most important etiological predisposing factors [4, 5]. Early diagnosis of this condition with prompt surgical treatment and early antibiotic therapy forms the cornerstone of its clinical management.

In a dedicated colorectal disease hospital, the majority of FG patients referred and managed have an anorectal origin that includes those primarily with perianal abscesses and anal fistulas. Here, infection from an anorectal focus often extends to the perineum and then to the scrotum, with some infections even spreading to the abdominal wall [2]. In our clinical practice of FG, however, some patients with a primary anorectal origin and involvement of the genital and perianal regions had sparing of the neighboring perineal soft tissues—a phenomenon not previously reported. It is postulated that this type of FG may represent a different process of tissue spread than the usual FG variant. The present study reviews our experience in the management of FG originating from the anorectal region, particularly in those without perineal involvement.

Patients and methods

Patients

This retrospective study was reviewed and approved by the Institutional Review Board of the Sixth Affiliated Hospital of Sun Yat-sen University, Guangzhou, China. All eligible patients with FG admitted to the Sixth Affiliated Hospital of Sun Yat-sen University and People's Hospital Affiliated to Fujian University of Traditional Chinese from May 2013 to April 2017 were included in the review. The inclusion criteria for this study were patients with a post-operative diagnosis of FG with an anorectal origin. A total of 63 patients with FG were initially identified, with 3 exclusions that were deemed not to have originated from the anorectal region.

Demographic and clinical variables

The patient demographic data, including age, body mass index (BMI), duration of symptoms and location of lesions, were obtained from hospital medical records. Any predisposing factors including diabetes mellitus, alcohol use, paraplegia/hemiplegia, chronic renal failure and epilepsy were recorded. BMI was calculated by dividing body weight (kg) with height squared (m^2) and obesity was defined as a BMI $> 30 \text{ kg}/m^2$. The duration of symptoms was defined as the interval from symptom onset to the first clinical evaluation in hospital. Clinical and laboratory data, including the body temperature, heart rate, respiratory rate and white blood count, were collected. Tachycardia was defined as >100 /minute with systolic hypotension defined as a blood pressure <90 mmHg and with tachypnea recorded when there was a respiration rate >20 /minute. The body surface area (BSA) involved by the infection was assessed using the burn index [6]. Imaging data (computed tomography [CT] and magnetic resonance imaging [MRI]) were also reviewed, along

with the operative characteristics and clinical patient outcomes.

Management

The general principles of FG management included hemodynamic stabilization and the intravenous administration of broad-spectrum antibiotics. Pus or necrotic tissue from the infected region was obtained for culture and sensitivity. Wide-spectrum antibiotic therapy was initiated empirically prior to the results of the microbial culture with modification of antibiotics when appropriate based upon culture sensitivities and the status of control of active infection. Tetanus antitoxin was routinely administered.

Following the correction of the fluid and electrolyte imbalance, a radical debridement was performed incorporating the soft tissues of the penis, scrotum, perineum and perianal regions as well as any other involved soft-tissue territory. Post-operatively, close glucose control was initiated and continued in patients with diabetes mellitus with daily wound irrigation using saline and povidone-iodine solution as well as hydrogen peroxide. Following debridement and the development of responsive granulation tissue, in those patients with scrotal skin loss involving $<50\%$ of the scrotal skin, the wounds were secondarily closed. In those patients with more advanced or even total scrotal skin loss, a delayed split-thickness skin graft (STSG) was used with the preferred use of a thicker non-meshed STSG as integumentary cover for penile defects.

Patients were contacted by telephone for a post-operative follow-up with direct questioning concerning wound healing, sphincter function and sexual function.

Statistical analysis

The SPSS version 13.0 (SPSS Inc., Chicago, Illinois) was used for analyses. Descriptive statistics included means and standard deviations or medians and ranges where indicated for continuous factors, as well as frequencies for categorical variables. The Chi-square or Fisher's exact test was used where appropriate for qualitative variables. A P -value <0.05 was considered statistically significant for all tests.

Results

Among the 60 included patients (56 males and 4 females), 32 (53.3%) were from the Sixth Affiliated Hospital of Sun Yat-sen University and 28 (46.7%) from Fujian Provincial People's Hospital (Figure 1). The mean age was 53.0 ± 15.9 years. The average BSA involved by the infection was $3.3 \pm 3.8\%$, with a mean BMI of $23.4 \pm 3.6 \text{ kg}/m^2$. The mean duration of symptoms prior to gangrene was 6.5 ± 3.7 days. The white blood cell counts and neutrophil granulocyte counts were markedly increased in 51 (85.0%) and 53 cases (88.3%), respectively. The predisposing factors and clinical manifestations are listed in Table 1.

Microorganisms were isolated on culture of the necrotic or purulent tissues in 34 patients while there was no necrotic tissue available for the remaining 26 cases. The common bacterial species isolated were *Escherichia coli* (55.9%) (Table 2). Only one patient (2.9%) had two organisms grown on the culture. Triple empirical antibiotics, including a third-generation cephalosporin, an aminoglycoside and metronidazole or clindamycin, were used in all the cases with a change in regimen in accordance with the findings of the sensitivity tests and continuance of antibiotic therapy until the active infection was controlled. The

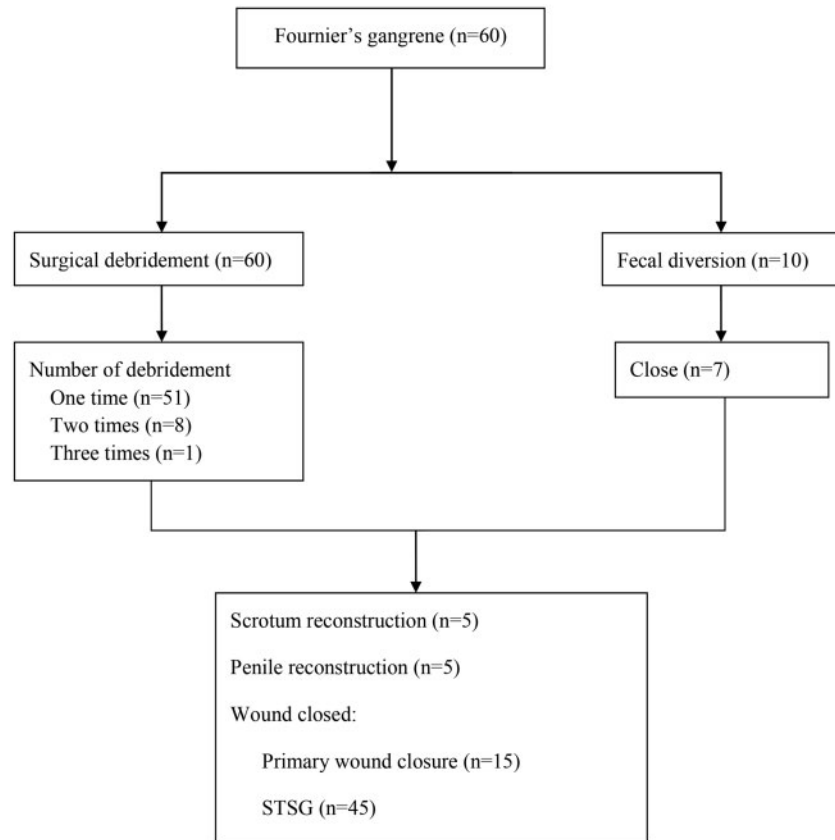


Figure 1. Participant enrolment and follow-up (flow diagram)

Table 1. Predisposing factors and clinical manifestations

	No. of patients (N = 60)
Predisposing factors	
Anal fistula	1 (1.7%)
Perianal abscess	42 (70.0%)
Diabetes mellitus	29 (48.3%)
Paraplegia/hemiplegia	2 (3.3%)
Chronic renal failure	4 (6.7%)
Epilepsy	0
Symptoms and signs	
Scrotal (labia majora) swelling	45 (75%)
Scrotal (labia majora) pain	41 (68.3%)
Scrotal (labia majora) redness	42 (70.0%)
Perineal pain	33 (55.0%)
Perinal pruritus	1 (1.7%)
Crepitus	28 (46.7%)
Fever greater 39°C	4 (6.7%)
Tachycardia	22 (36.7%)
Systolic hypotension	0
Tachypnea	15 (25.0%)

mean duration of use of broad-spectrum antibiotics was 15.7 ± 8.5 days (range 6–38 days).

CT scans were performed in 49 cases and MRI was conducted in 11 patients. Based upon the imaging data and clinical manifestations, involvement of the genital and perirectal regions was detected in 52 and 59 cases, respectively, while the perineum was specifically not involved in 7 (11.7%) patients. An abscess in the ischio-rectal fossa was evident in 47 cases (78.3%).

Table 2. Pathogen culture of the necrotic or purulent tissues

Microorganism	No. of patients (N = 34)
<i>Escherichia coli</i>	19 (55.9%)
<i>Enterococcus faecalis</i>	1 (2.9%)
<i>Proteus mirabilis</i>	1 (2.9%)
<i>Klebsiella pneumoniae</i>	1 (2.9%)
<i>Peptostreptococcus</i>	1 (2.9%)
<i>Pseudomonas aeruginosa</i>	1 (2.9%)

Table 3. Location of lesions

Location	No. of patients (N = 60)
Penile shaft	22 (36.7%)
Scrotum (labia majora)	52 (86.7%)
Perineum	53 (88.3%)
Anorectum	59 (98.3%)
Inguinal	29 (48.3%)
Thigh	14 (23.3%)
Suprlevator	18 (30.0%)
Ischio-rectal fossa	47 (78.3%)
Anterior abdominal wall	18 (30.0%)
Posterior pararenal space	1 (1.7%)

Eighteen (30.0%) patients showed a suprlevator abscess extension linked to the ischio-rectal fossa via translevator passage. The locations of ancillary lesions are listed in Table 3. When the internal inguinal ring was dissected in those seven cases in which the perineum was unaffected, purulent fluid was able to

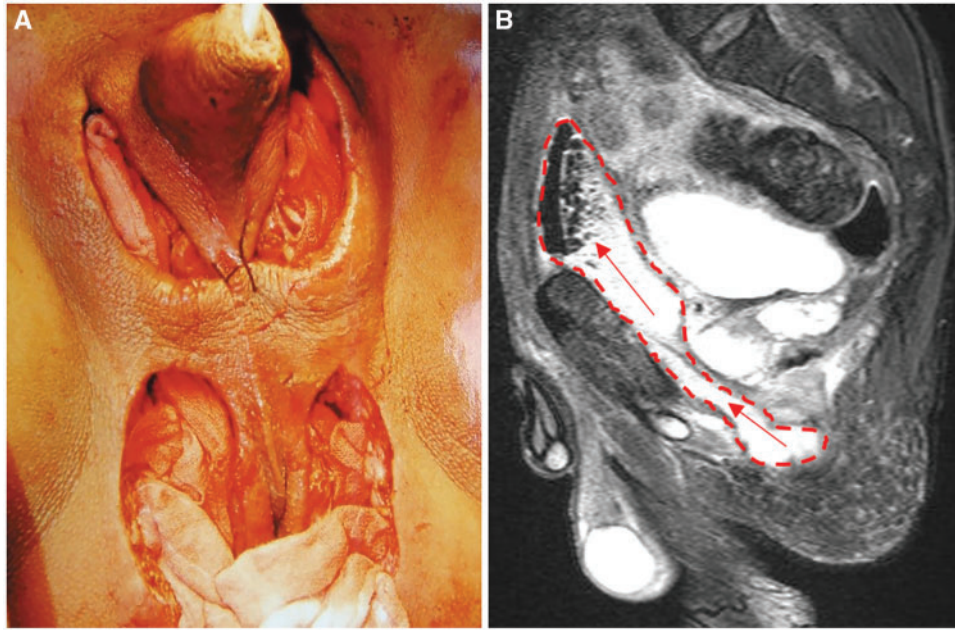


Figure 2. Fournier gangrene without perineal involvement. (A) Representative surgical image of genital and anorectal involvement with relative perineal sparing. (B) The MRI showed that infection involved the perianal region and spread to the suprapleator space and reach retropubic space (red arrows), but spared most of perineal tissue.

be expressed from the proximal end of the inguinal canal where, on exploration, there was a direct communication along the canal into the retropubic space. Although the genital region was involved in all seven cases, the severity of the infection locally was minor, with less purulent fluid emanating from the distal end of the inguinal canal when compared with that found at its proximal end. In each of these cases, the skin, fascia and most of the perineal musculature were unaffected (Figure 2).

The treatment, including surgery and antibiotic therapy, was generally effective. However, several major complications, including one case of generalized sepsis, one fungal infection, one massive hemorrhage from erosion of an abdominal wall vessel and one septic death, were identified in this cohort. No patient underwent an orchidectomy and required urinary diversion. Ten patients (16.7%) needed a protective colostomy. Vacuum-assisted drainage (VSD) was used in two cases only in our clinical practice. One patient with a colostomy died of delayed sepsis, while all the patients who did not undergo diversion survived; the mortality rate is similar between patients with and without a colostomy (10 vs 0%, $P = 0.167$). The median interval between the debridement procedure and colostomy closure was 7 months (range 4–12 months). In addition, in this series of all recruited patients, secondary debridement was performed in eight cases (13.3%), with one (1.7%) undergoing debridement on three occasions.

The mean time to oral intake was 4.4 ± 4.6 days and no ileus was observed in the present study. Reconstruction of the scrotum was performed in five cases, with the average time between initial debridement and penoscrotal reconstruction being recorded as 25.3 ± 5.6 days. The wounds were directly closed in 15 cases (25.0%) and a STSG was used in the remaining 45 patients (75.0%). Thinner, durable non-meshed STSG was used in five patients specifically for penile reconstruction. The average time until STSG was 26 days (range 17–64 days).

Follow-up was achieved in 50 patients (83.3%), with a mean follow-up period of 26.9 ± 14.1 months. All patients were in good health at the time of their most recent follow-up.

Discussion

FG is a rare but frequently fatal disease characterized by acute necrotizing fasciitis that develops in the perineum and the external genitalia. Typically, a rapid clinical progression occurs with a mixed aerobic and anaerobic bacterial infection. The present study demonstrated that FG originating in the anorectal region was rapidly progressive and life-threatening. Early diagnosis and aggressive surgical debridement are the mainstays of FG management. A possible route of infection for an anorectal origin can be identified with CT imaging. Further, a protective colostomy for anorectal-derived FG is not mandatory.

In this study, we found several common causative microorganisms, such as *E. coli*, where most bacteria originated from the bowel even though the bacterial profiles of non-anorectal-derived and anorectal-derived FG are similar [7, 8]. The perceived rate of spread of fascial necrosis caused by mixed organisms has been reported to be as high as 2.536 cm per hour [4], sometimes with a rapid progression to systemic toxicity. The treatment should involve a broad-spectrum antibiotic therapy with, in our opinion, a focus on combating Gram-negative bacteria of gastrointestinal origin. This is accompanied by a timely and radical debridement emergently performed so as to stop the progression of the infection.

Although the diagnosis of FG is often made clinically, early diagnosis can be achieved with a more accurate assessment of the disease extent using emergency CT and MRI, which can also direct the areas of infection that need debridement. An advantage is that the communications between spaces are able to be assessed by reconstructed imagery defining infections spreading along fascial planes along the perineum and beyond. In cases of FG originating from perirectal sepsis, infection

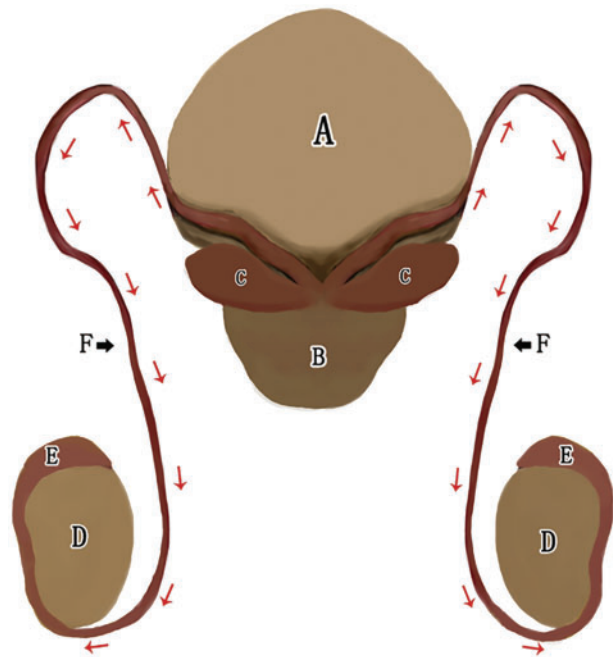


Figure 3. Postulated pathogenetic mechanism of the spread of infection that could involve the perineal region and spread across the levator plate to the supralelevator space and then subsequently reach the retropubic space and the inguinal region whilst sparing most of the perineal soft tissues. (A) Bladder; (B) prostate; (C) seminal vesicle; (D) testicle; (E) epididymis; (F) inguinal canal.

frequently spreads along Colles' fascia and progresses anteriorly along the Dartos fascia to involve the scrotum and the penis [9]. However, in seven cases (12%) in our series, the genital and perirectal regions were involved but the perineum was spared, as confirmed by clinical, radiological concordance and at surgical exploration.

These findings are at variance with the current literature concerning the spread of infection in FG, where it has been suggested that infection can not only spread along fascial planes, but also extend deeply into muscle [10]. Such myonecrosis is usually caused by Clostridial species, although it can occasionally result from virulent *E. coli* infection [11]. In this event with *E. coli* culture, it is advised to consider alternative pathways for the progression of local sepsis—a state best defined by CT and MR imaging. Infection with a particularly virulent microorganism or, more commonly, from a combination of microorganisms originating from the perirectal region may also extend as an abscess across the levator plate into the supralelevator space, as shown with specialist scanning in 18 cases (30%) in our present study. This demonstrates that mesorectal and seminal vesicular involvement permits spread along the ductus deferens to involve the inguinal region and the scrotum (Figure 3). Further infection can pass superiorly in this plane along Scarpa's fascia to the anterior abdominal wall (30% in our series) whereas supralelevator sepsis can involve both the retrorectal and retroperitoneal spaces where, on CT scan in one of our cases, there was a large focus of gas evident in the posterior aspect of the pararenal space. Although CT and MRI have great specificity in the evaluation of the disease extent in FG [12], they can potentially miss the type of perineal sepsis seen in our study in which the false negative rate when compared with surgical exploration was 11.7% (seven cases).

In the present study, no patients required an orchidectomy, although the orchidectomy rates in FG have been reported to be

up to 25% in some series [13]. This reflects the different origins of blood supply where the testicular artery is a direct branch from the aorta and where the blood supply to the scrotum comes from the pudendal arterial branches of the femoral artery [14]. Once the infection involves the testis, however, this may be indicative of retroperitoneal or intra-abdominal sepsis [15]. In our series, all patients underwent timely and radical debridement, which is an essential part of FG management, with liberal use of the second-look procedure. In this regard, eight patients (13.3%) underwent a second debridement, with one patient (1.7%) undergoing debridement three times. It would seem that the VSD would be specifically beneficial, relieving post-operative pain and shortening the length of hospital stay. Future cases will be managed with the VSD in an effort to determine its clinical value [7].

It still remains controversial whether fecal diversion is needed in patients presenting with FG. Some physicians prefer to perform a routine fecal diversion in these patients in order to decrease wound contamination and to promote faster wound healing [16, 17]. In this respect, Chen and colleagues have reported that, in FG patients with anorectal disease, those undergoing a primary diverting colostomy have a lower mortality rate overall when compared with those undergoing secondary colostomy treatment [18]. This issue is complex, since mortality may better correlate with the initial extent of disease, resulting in a bias of colostomy being associated with worse prognosis cases. Concerning this point, Korkut et al. [19], in a retrospective comparative study, reported that the mortality of patients not needing a stoma was low (7%), but increased to 38% when a stoma was required. It should be remembered that serious stoma-related complications can occur, including wound infection, stoma ischemia, prolonged ileus and evisceration, suggesting that the decision for diversion should be measured (16.7% in this study). In our opinion, a protective colostomy is advisable in order to prevent fecal contamination when there is attendant sphincter dysfunction or involvement, rectal perforation and accompanying immunodeficiency.

In conclusion, FG originating in the anorectal region may be rapidly progressive and life-threatening. The infection may spread superiorly to the genital region without involvement of the perineal soft tissue. An aggressive surgical debridement of all non-viable tissue is essential for successful management of the disease. In our view, a protective colostomy is not mandatory.

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Conflict of interest

None declared.

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