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Case Report

Fournier Gangrene as a Manifestation of Undiagnosed Metastatic Perforated Colorectal Cancer

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Fournier gangrene is a necrotizing soft tissue infection involving the perineum. We present a case of Fournier gangrene as the clinical presentation of perforated metastatic rectal cancer. The patient is a 78-year-old man in a nursing home who presented to our institution with necrosis and ischemia of the scrotum. After wide debridement of necrotic tissue and bilateral orchiectomy, computed tomography was carried out to investigate abnormal findings seen on his chest X-ray, which revealed multiple pulmonary metastases as well as a mass highly suspicious for a perforated rectal mass. Once stable, a diverting colostomy and biopsies of the rectal mass were performed, confirming the presence of a metastatic, poorly differentiated rectal adenocarcinoma. Albeit an unusual etiology of Fournier gangrene, this case highlights the rare but important causes of this deadly condition and teaches us to be cognizant of the variations in the presentation of colorectal cancer.

Key words: Fournier gangrene – Necrotizing fasciitis – Colorectal cancer – Perforated colorectal cancer – Metastatic colorectal cancer

Fournier gangrene (FG) is a rare, life-threatening necrotizing fasciitis that usually involves the perineal or genital areas. Prompt identification of this deadly disease is fundamental for appropriate treatment. Knowledge of the common predisposing factors and conditions aids in early diagnosis. Frequent etiologies include trauma, surgical wounds, and in rare instances, malignancies.

We present a case of an undiagnosed, perforated metastatic rectal carcinoma presenting as FG.

Case Report

A 78-year-old man was transferred from a nursing home with worsening infection and necrosis of the perineal and scrotal areas. The infection had persisted over the course of 5 days, until the patient had expressed his discomfort. At this point, the patient was transferred to our services for evaluation and management.

His past medical and surgical history included hypertension, history of a cerebrovascular accident,

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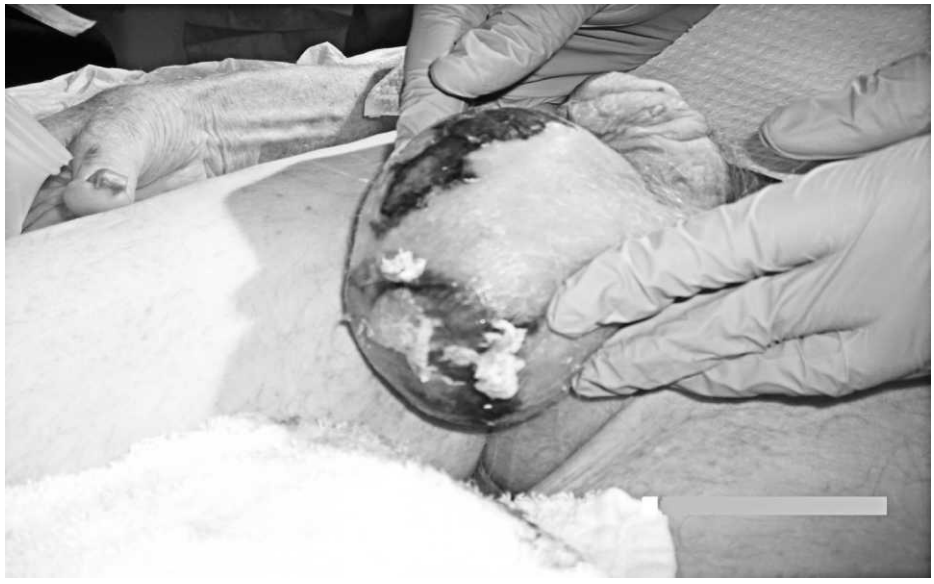

Fig. 1 Further evaluation of scrotum with significant necrosis noted.

dyslipidemia, and coronary artery disease status after coronary stenting. He was on several antihypertensive medications, including metoprolol, enalapril, and hydrochlorothiazide, along with a low-dose aspirin and atorvastatin. The patient also had a 30 pack-per-year smoking history.

The patient was alert, conversant, and appropriate. No signs of shock were apparent based on clinical exam. However, he was noted to have an extensive area of necrosis along with crepitus noted within and around the scrotum. This extended upward along the inferior aspect of the penis. Marked erythema and induration were also noted (Fig. 1). Rectal examination revealed exquisite tenderness around the anus, but no discernible mass was felt.

Initial laboratory investigations revealed significant leukocytosis of 36,800 per cubic millimeter, a hematocrit of 0.274, and a potassium of 2.3 mmol/L. The remainder of the patient's laboratory investigations were within normal limits. This translated to an FG Severity Index (FGSI) score of 8, which correlates with an approximately 5% in-hospital mortality rate.[1] A preoperative chest x-ray (CXR) showed suspicious nodules noted in both the right and left lung (Fig. 2). However, given the patient's clinical exam and laboratory results, he was taken urgently to the operating room.

The patient was started on gentamicin, vancomycin, and metronidazole empirically. Urology Services was consulted prior to surgery, and a joint operation was carried out. Inspection revealed a large retroperitoneal abscess, which was drained via the perineum. Debridement of necrotic scrotal tissue, bilateral orchiectomies, and excision of necrotic tissue from the base of the penis and surrounding perineum were also done (Fig. 3). Wound and abscess cultures were taken for culture and sensitivity. These later revealed *Escherichia coli*; thus, cefepime was added to his antibiotic regimen. The patient was then transferred to the surgical intensive care unit for postoperative resuscitation.

While the patient was in the intensive care unit, we proceeded to investigate the underlying etiol-

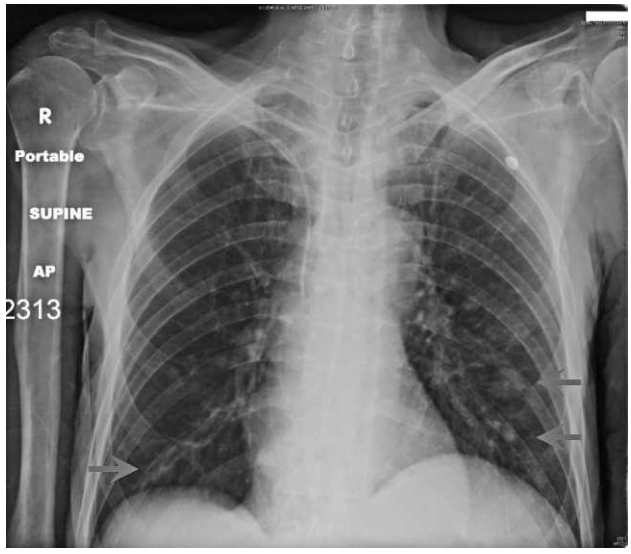

Fig. 2 Preoperative CXR showing multiple lung nodules.

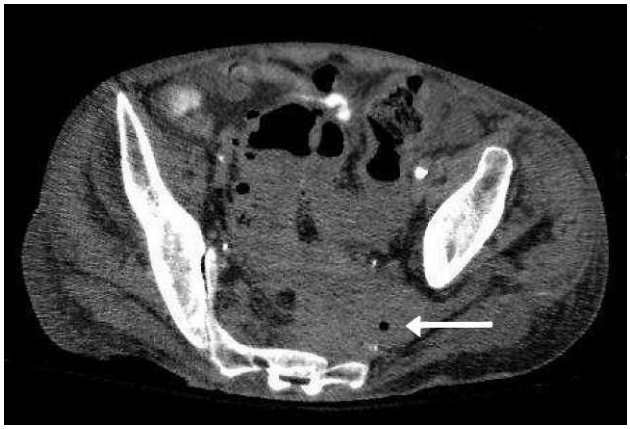

Fig. 3 CT showing extraluminal air adjacent to rectal mass.

ogy of the patient's FG. Furthermore, workup regarding the suspicious lung nodules was carried out. This included a carcinoembryonic antigen level, which was elevated at 70.2 ng/mL. A computed tomography (CT) scan revealed increased soft tissue density in the left ischiorectal fossa extending to the sciatic notch and adjacent sacrum. Additionally, air bubbles were noted in the left groin and along the base of the penis. This was concerning as the air bubbles were present likely as a result of perforation from a rectal neoplasm (Figs. 3–5). Finally, multiple nodules were seen in both lungs, the largest measuring 10 mm (Fig. 6). Based on the CT scan findings, a diagnosis of perforated metastatic rectal cancer was highly suspected.

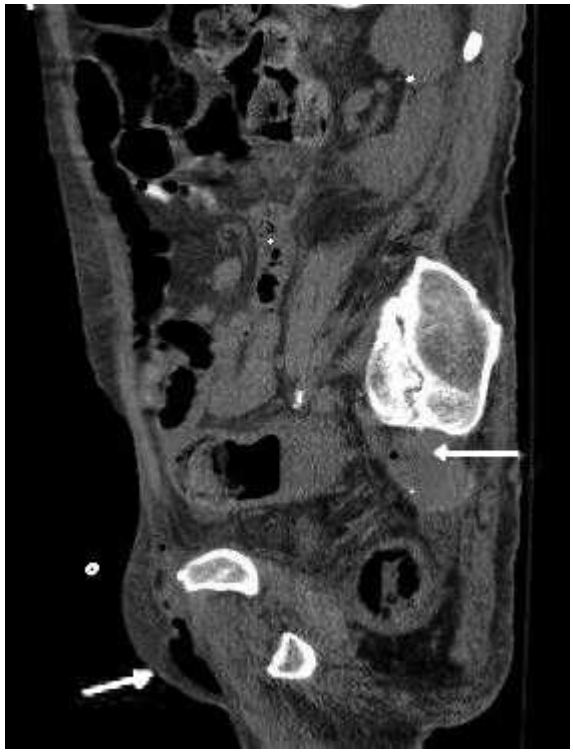

Fig. 4 CT sagittal view showing perineal necrosis and extraluminal air.

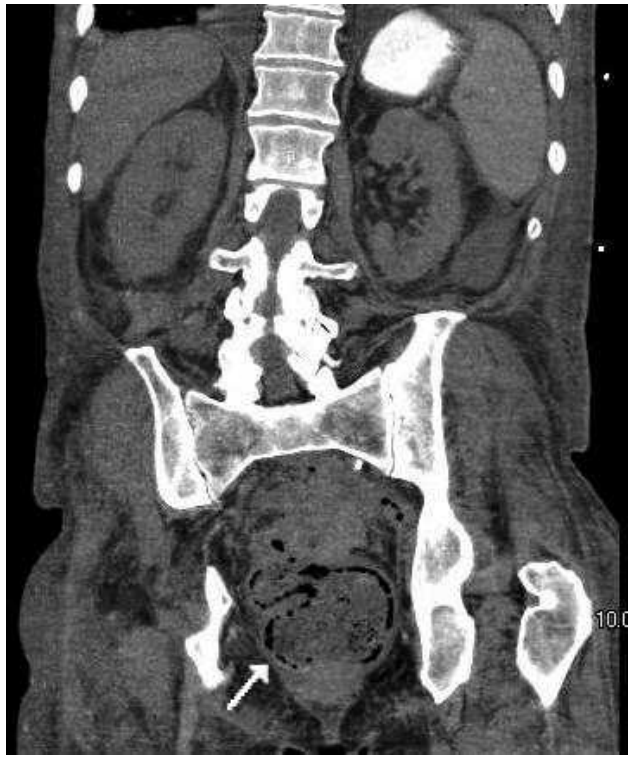

Fig. 5 CT coronal view of rectal mucosal ischemia and extraluminal air.

Five days after the initial operation and upon adequate resuscitation, the patient was brought back to the operating room for a diverting colostomy and a rigid proctoscopy to obtain tissue samples of the mass noted in the rectum. Approximately 600 mL of serous peritoneal fluid was noted upon entry into the peritoneal cavity. This was sent for cytology, but it failed to reveal any malignant cells. Rigid proctoscopy revealed a large rectal mass 10 cm from the anal verge, which was biopsied. Final pathology revealed a moderately to poorly differentiated adenocarcinoma of the rectum with a small focus of lymphovascular invasion; no perineural invasion was seen.

The patient did well initially following surgery. However, it was clear that his overall prognosis was poor, and this was discussed.

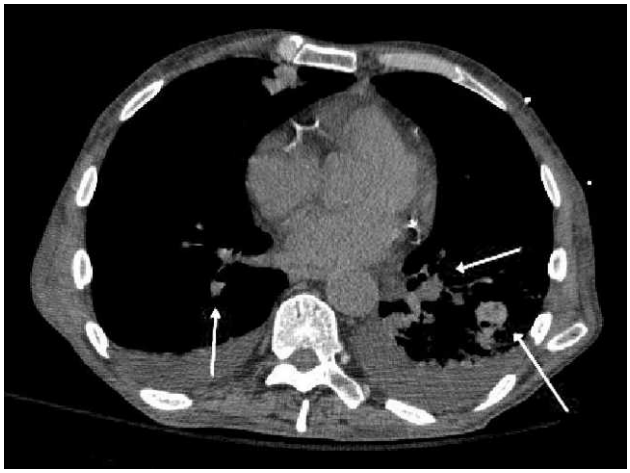

Fig. 6 CT axial view revealing multiple bilateral pulmonary nodules.

Discussion

FG was first described in 1764 by Baurienne. However, the condition was named after the French dermatologist and venereologist Jean-Alfred Fournier, following his presentation of 5 cases in 1883. In this series, Fournier described a rapidly progressive gangrene of the penis and scrotum in 5 previously healthy young men.[2] The modern-day term now refers to a polymicrobial necrotizing soft tissue infection involving the superficial and deep fascial planes of the genital, perineal, or perianal areas. With fewer than 2000 cases reported in the literature,[3,4] FG is a rare infection, occurring primarily in elderly men in their sixties and seventies. FG has an average incidence of 97 cases per year[4] and a prevalence of approximately 1 case in 7500 persons.[5] Men are 10 times more likely to develop this infection, because women have better drainage of the perineal region through vaginal secretions.

Originally, FG was thought to be idiopathic, but an identifiable cause can now be identified in up to 95% of cases.[6] The infection can be secondary to anorectal, urogenital, trauma, or dermatologic pathology, in which a mixture of both anaerobic and aerobic bacteria may be inoculated.[7,8] The infectious agent is most often polymicrobial (62%), with *Streptococcus* spp (31%), *Bacteroides* spp (27%), and *E coli* (24%) being the most common isolates.[9]

Comorbid conditions can also predispose a patient to the development of FG, with diabetes mellitus being the most common factor (55.6% of cases[10]). Other factors include morbid obesity, vascular diseases, and malignancies.[11,12] In rare cases, FG may be secondary to an occult colorectal malignancy, which has been reported only several times since FG was first described.[13–20] Apart from gross inspection of the involved tissues, pathologic analysis also plays a role in diagnosis.

Extensive studies of pathologic specimens from individuals with FG have characteristic findings. Pathognomonic findings based on pathologic evaluation of involved tissue include necrosis of superficial and deep fascial planes, fibrinoid coagulation of the nutrient arterioles, polymorphonuclear cell infiltration, and microorganisms identified within the involved tissues (Figs. 7 and 8).[21] Regardless, clinical examination remains the most reliable means of identification of this disease. Blood tests, radiographic imaging, and pathologic sampling are only adjuncts to diagnosis and are primarily used in determining the severity of the condition.

Treatment with broad-spectrum antibiotic coverage to include staphylococci, streptococci, Enterobacteriaceae, and anaerobes should be promptly initiated. However, aggressive surgical management remains the mainstay of treatment. This includes wide excision of all necrotic tissue down to bleeding tissues.[22] Oftentimes repeated operative debridements may be needed to sufficiently debride all involved tissue. Chawla *et al*[23] showed that repeated debridements often correlated negatively with sur-

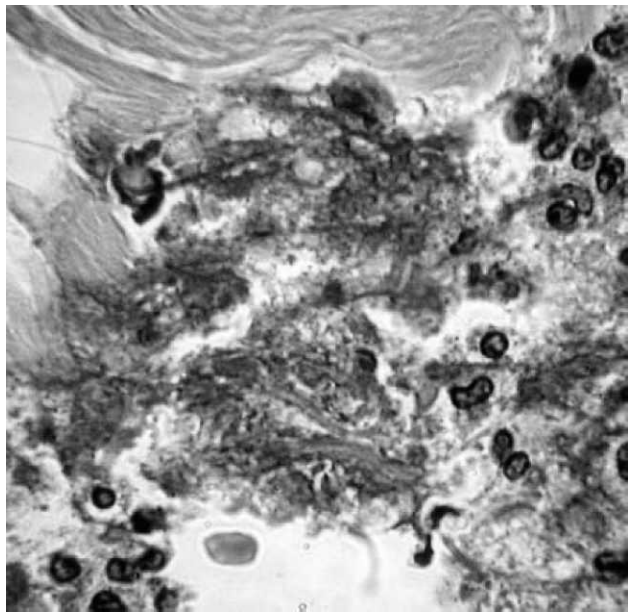

Fig. 7 Photomicrograph of FG showing acute inflammatory cells in necrotic tissue (oil immersion ×1000).[21]

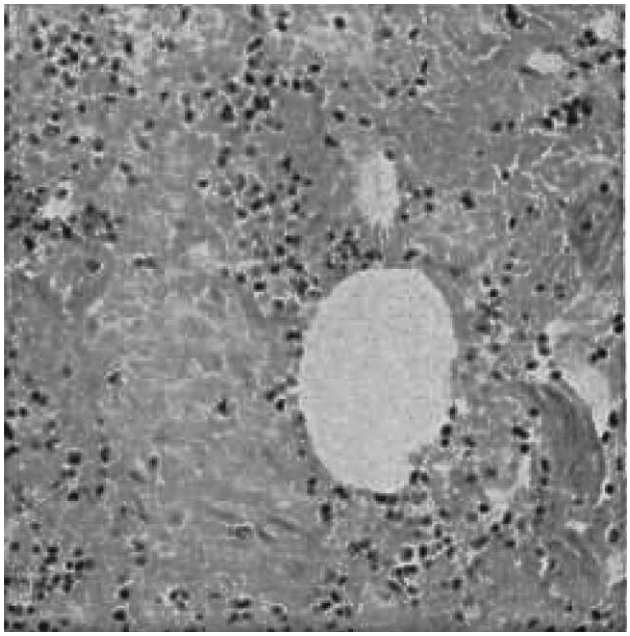

Fig. 8 Photomicrograph showing gas-filled pockets present within fibrous tissue, infiltrated by erythrocytes and neutrophils (hematoxylin and eosin, 1000× = high power).[22]

vival, speculating that these patients generally had more extensive disease, were in poorer health at baseline, and had progression to systemic sepsis, despite early aggressive surgical intervention.

In extensive perineal cases, fecal diversion may be necessary to allow for adequate wound healing. Additionally, formal reconstruction with skin grafting or flap closure may be required if substantial skin loss is noted. Other treatment modalities such as hyperbaric oxygen therapy[24] and topical unprocessed honey[25] have been tried, but they remain investigational. Unfortunately, despite aggressive therapy mortality remains high at approximately 40%, and nearly 80% if sepsis is present on initial presentation.[26]

Given the acuity of this condition, only retrospective reviews have been published. Therefore, drawing prognostic information from these studies is relatively unreliable. As a result, Laor *et al*[1] developed the FGSI in 1995, which assesses 9 clinical parameters and the extent of deviation from normal. They determined that a score of 9 or higher combined with advanced age correlated with increased mortality.[1] Later, Corcoran *et al*[8] calculated that an FGSI score lower than 9 corresponded to a mortality of 4%, whereas a score higher than 9 had a 46% mortality rate. Additionally, an anorectal source, advanced age, extensive disease, shock or sepsis on initial presentation, renal failure, and hepatic dysfunction correlated with a higher mortality.[27] Death was usually the result of systemic illness, such as multiple organ failure, coagulopathy, or sepsis. One study evaluated the usefulness of lactate level in predicting overall clinical outcome and found that a level greater than 4.0 mmol/L was an independent predictor of mortality.[28] On the contrary, factors associated with an improved prognosis include localized clinical disease, absence of systemic toxicity (*e.g.*, low FGSI), and age younger than 60 years.[29]

FG remains a condition with both a high morbidity and high mortality. Although a rare entity, having an understanding of the common predisposing conditions along with the causative agents allows prompt diagnosis. Institution of broad-spectrum antibiotic coverage is important. However, early and aggressive surgical debridement remains fundamental in treatment. This case highlights an unusual cause of FG and points out that identifying the underlying condition is sometimes essential in overall control of this deadly disease. Therefore, one should be cognizant not only of the more common conditions, but also of the unusual and atypical ones, because this may ultimately dictate control of the offending source. It was clear in this case that this patient's infection would have persisted had the perforated rectal cancer been undetected. Unfortunately, because of the metastatic nature of his disease, local control and fecal diversion were purely palliative.

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