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Fournier's gangrene as the unveiling manifestation of occult perforated diverticulitis

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Abstract
We present a case of a 44-year-old Caucasian Male patient with no known identifiable risk factors for the development of Fournier's gangrene who presented in septic shock with clinically significant scrotal pain, and rapidly progressive erythema and edema. The patient was resuscitated and treated in accordance with the surviving sepsis guidelines immediately prior to operative management. Consequently, the patient underwent serial, extensive, excisional debridement to adequately manage this life-threatening, infectious pathology. The diagnostic laparoscopy revealed intraabdominal adhesions, which, although not immediately conclusive, retrospectively informed clinical suspicion of an underlying process such as perforated diverticulitis as the etiology of his presentation. This prompted further postoperative evaluation, and ultimately a Hartmann's procedure for definitive source control. This report highlights the need for prompt recognition and aggressive management of life-threatening necrotizing soft tissue infection (NSTI), and rare complications associated with undiagnosed or unsuspected perforated diverticulitis.

Keywords: Excisional debridement, Fournier's gangrene, Hartmann's procedure, necrotizing fasciitis, perforated diverticulitis

Introduction
Fournier's gangrene is a rare form of necrotizing fasciitis, which is a rapidly progressive, severe inflammatory and infectious disease process that requires prompt recognition and timely operative management if diagnosed [1]. Diagnosis is often delayed, as early clinical findings may be nonspecific [2]. While careful review of initial imaging is important - particularly if manifestations such as crepitus are absent - it should never postpone timely surgical intervention when clinical concern is high. Commonly, Fournier's gangrene is seen in male patients with diabetes or those in immunocompromised states [3]. In the original descriptions describing this pathology, the disease arose in healthy subjects without an identifiable cause, but more recent literature attributes this process to the aforementioned epidemiological factors as well as those arising from urologic or colorectal origin [4]. The rates of colorectal disease resulting in presentation with Fournier's gangrene is not definitively quantified [4] but is a known rare cause. We present a rare case whereby a patient developed Fournier's gangrene in the setting of occult, perforated diverticulitis necessitating aggressive surgical debridement, Surgical Intensive Care Unit (SICU) admission, and a Hartmann's procedure for definitive source control.

Case Report
A 44-year-old male patient with a complicated history significant for deep venous thrombosis (DVT), pulmonary embolism (PE) status post inferior vena cava (IVC) filter placement, previously on Xarelto, asthma, Crohn's disease and an appendectomy presented to the emergency department with clinical findings most suggestive of Fournier's gangrene.
The patient initially reported experiencing significant pain in his right testicle approximately thirty-six hours prior to initial presentation. This pain was associated with rapidly progressive erythema and scrotal edema prompting the desire to seek urgent evaluation. Notably weeks preceding the patient's current presentation, he mentioned having experienced several upper respiratory infections (URI), including coronavirus disease (COVID) and influenza A, for which he was prescribed prednisone monotherapy - a known risk factor for developing NSTIs due to its immunosuppressive effects.
Upon arrival at the emergency department (ED), the patient was notably in septic shock.

The patient was expeditious and appropriately provided 30 cc/kg of crystalloid fluid resuscitation and broad-spectrum antibiotics-including Vancomycin, Zosyn and Clindamycin-were initiated in accordance with surviving sepsis guidelines [8]. The patient underwent STAT imaging that included a computed tomography of the abdomen and pelvis (CT A/P) which not only demonstrated extensive inflammatory and gas forming changes in the pelvis and scrotum extending alongside the right spermatic cord into the anterior right abdominal soft tissues consistent with Fournier's gangrene, but also suspicious intra-abdominal collections alongside the urinary bladder, and evidence of suspected diverticular disease (Figure 1). Significant labs included an elevated leukocytosis of 15.8, c-reactive protein 25.7, hemoglobin 12.2, sodium 129, creatinine 0.87, glucose 121. Laboratory risk indicator for necrotizing fasciitis (LRINEC) was calculated and a score 8 was defined. Urology was consulted due to concern of extensive involvement of the right testicle. The patient was emergently transported to the operating room whereby he underwent a cystoscopy, scrotal debridement with Urology and further excisional debridement of the bilateral groins and scrotum, and a diagnostic laparoscopy with the Acute Care surgeon under general anesthesia. Intraoperative findings included a necrotic scrotum that involved the subcutaneous and fascial tissues diffusely throughout the scrotum, no involvement of the corporal cavernosal tissue, urethra or testicles, necrotic tissue to the level of the right anterior superior iliac spine (ASIS) and perineum, dishwater-like fluid diffuse throughout the operative site, significant intra-abdominal adhesions to the anterior lower abdominal wall and pelvis limiting complete evaluation of the intra-abdominal contents, serous and bloody ascites, a normal appearing descending colon and although incomplete visualization of the sigmoid colon - that which was seen also appeared normal. All necrotic tissue described was excised via a combination of sharp dissection and electrocautery. Intraoperative wound cultures obtained. Patient remained intubated postoperatively and was promptly transported to the SICU for further management of septic shock on pressor support.

On postoperative day (POD) 1, resuscitation and appropriate antibiotic therapy continued, and a second look operation was scheduled to take place. Intraoperatively, the dressings were replaced, testicles redressed with petroleum gauze and Penrose drains placed through pre-existing counter incisions. Final wound dimensions measured 39x6x4 cm on the right, and 30x6x4 cm on the left. Both measurements included undermining. Patient was extubated and transported back to the SICU.

Attempting to elucidate the etiology of the patient's disease process, and due to adhesions identified upon initial laparoscopy within the index operation, a repeat CT A/P with triple contrast was obtained (Figure 2). This study demonstrated a communication of the sigmoid colon with pelvic abscesses suggestive of perforated diverticular disease. Consequently, the patient returned to the operating room three additional times for excisional debridement and dressing changes prior to undergoing an exploratory laparotomy and Hartmann's for management of the patient's perforated diverticulitis that preceded the development of the NSTI. This procedure was successfully completed but was complicated by an estimated blood loss (EBL) of two liters and hypotension requiring further resuscitative management within the SICU. Plastic Surgery was consulted during the patient's inpatient admission, and on hospital day (HOD) 14, the patient underwent successful closure of the complex, large perineal defect.

The patient's postoperative course was further complicated by hypoxia, tachycardia, right back and flank pain, and a Jackson-Pratt (JP) drain with persistently voluminous output. Appropriate imaging studies obtained included CT angiogram (CTA) to evaluate for pulmonary embolism (PE), and rectal stump leak - both of which were negative. Drain studies that included creatinine, amylase and triglycerides were negative. Findings suggestive of possible pneumonia were adequately treated with antibiotic therapy.

The patient ultimately progressed well, functional status appropriate for discharge home, on antibiotics and with the follow up for drain care and outpatient colonoscopy.

After discharge, the patient underwent colonoscopy and planned an elective Hartmann's reversal that occurred one hundred and fifty-four days after his initial presentation. This procedure was tolerated well, but the postoperative course was complicated by CT evidence of acute non-occlusive DVTs of the bilateral internal iliac, and femoral veins. Hematology consulted. Patient was discharged on Xarelto with plans to follow for further anticoagulation guidance. The patient was then discharged on POD 6 and has been progressing well since.

Discussion

Fournier's gangrene is an infection that commonly involves the underlying soft tissue and or fascial planes[1]. It tends to affect immunocompromised patients but can affect those that are healthy as well [1]. Characteristically, this rare infection has a strong male predilection with a male to female ratio of 10:1, although women have demonstrated higher morbidity and or mortality rates[1]. Both aerobic and anaerobic bacteria are typically identified within cultures obtained from these wounds [1]. Gram positive organisms often seen include group A Streptococcus and Staphylococcus aureus [1]. Gram-negative organisms such as Escherichia coli and Pseudomonas aeruginosa may be present in wound cultures as well [1].

Clinical identification of this disease is fraught with many challenges and often relies on appropriate radiographic imaging studies for diagnosis. This may be a result of varying etiologies that might include diabetes mellitus, urinary tract infections, infections of the perineal region, intra-abdominal or lesions as common as pimples[1]. Rapid identification may also be a consequence of minimal or no obvious physical manifestations, as well as generalizable symptoms consequently providing the infection an opportunity to progress [1]. To assist in diagnosis, validated scoring systems such as the LRINEC score incorporate key clinical and laboratory variables. However, neither imaging nor these scoring tools offer definitive diagnostic certainty and should not supersede a high clinical index of suspicion.

The symptoms experienced by this patient included increasing scrotal pain, erythema and edema that began approximately thirty-six hours prior to initial presentation. The patient presented in septic shock, and with clinical and laboratory findings suggestive of a rapidly progressive infectious process. The CT A/P obtained further demonstrated findings indicative of an NSTI or Fournier's gangrene (Figure1). Identification and treatment with crystalloid, broad spectrum antibiotics and emergent surgical debridement of all devitalized tissue is warranted in all cases of necrotizing fasciitis. Ultimately, patients often require serial debridement, prolonged ICU admissions and antibiotics until definitive source control has been obtained.

The patient presented required serial debridement, as well as a Hartmann's procedure, and plastic surgery intervention for definitive management of his conditions. This patient's etiopathogenesis was ultimately attributed to perforated sigmoid diverticulitis with direct extension of polymicrobial contents to the inguinal and perineal regions. Although this patient did not exhibit some of the more common causes of this condition, the previously prescribed course of steroids for a URI may have suppressed their immune function, with a superimposed episode of perforated diverticulitis triggering a cascade of complications that ultimately led to the development of this patient's disease.

Perforated diverticulitis uncommonly presents as the etiology for this pathologic disease process despite it being common with a prevalence ranging from 65% of those aged 85 years, 30% of those aged 60 years and approximately less than 5% in those aged 40 or younger [2]. Despite this, there are accounts of patients suffering from an NSTI because of this. One such report described by Henry D.I. De'Ath *et al*. described a patient with sigmoid perforation that presented with pneumoscrotum. Despite the clinically apparent physical manifestations of an underlying disease process, the patient underwent a chest plain film, gastrografin swallow, and an CT A/P[7]. Ultimately, this patient underwent an emergent Hartmann's procedure, but unfortunately expired on the operating table [7].

In the evaluation of patients with clinically ambiguous pathologies, prompt diagnostic imaging serves a critical role in the initial workup. However, effective management also relies heavily on operative planning. In the case presented, the diagnostic laparoscopy performed afforded the identification of intra-abdominal adhesions raising suspicion for the etiopathogenesis of this disease process. We present this case to provide an example of a rare clinical scenario in which perforated sigmoid diverticulitis preceded the development of Fournier's gangrene. When formulating a differential diagnosis for this pathology in patients lacking typical predisposing comorbidities, it is critical to consider more common diseases, such as diverticulitis, as potential sources of such conditions.

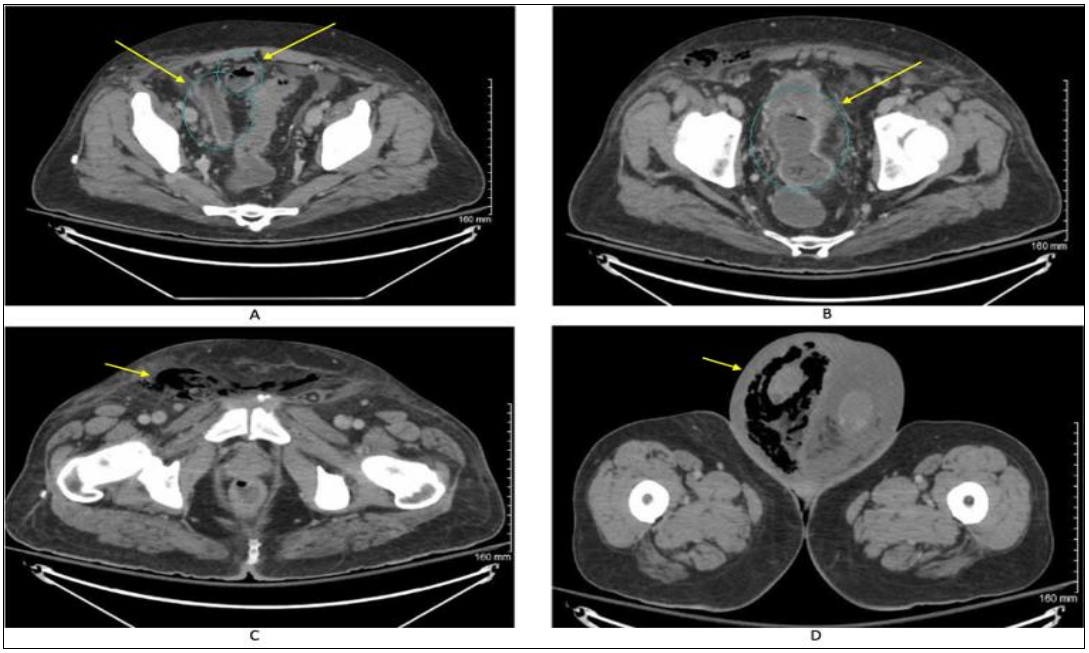

Fig 1 (A-D): (CT A/P axial image; cranial to caudal): Extensive inflammatory changes identified within the pelvis and scrotum. Fluid collections identified within the pelvis which do not definitively correspond with fluid-filled bowel loops, concerning abscess. Gas-forming infection involving the right hemiscrotum and scrotal soft tissues, extending up along the right spermatic cord in the anterior right abdominal soft tissues.

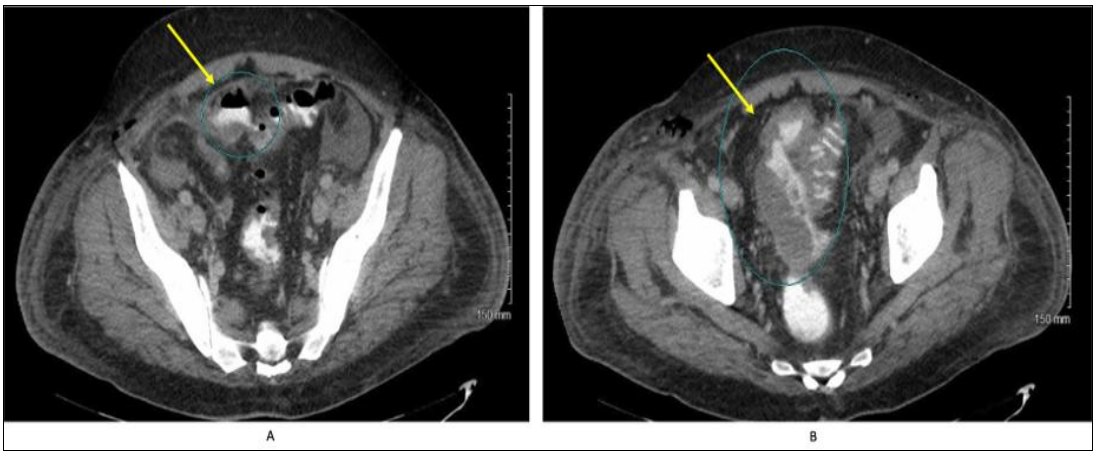

Fig 2 (A-B)(CT A/P axial image; cranial to caudal): Abscess with enteric communication. Tubular fluid collection extends along the anterior midline pelvis, then courses posteriorly into the prerectal space. This is likely intimate with a diseased loop of sigmoid colon. Findings most consistent with diverticular abscess.

Abbreviations
Abdominal - ABD
Anterior Superior Iliac Spine - ASIS
Computed Tomography Abdomen and Pelvis - CT A/P
Computed Tomography Angiography - CTA
Coronavirus Disease - COVID
Deep Venous Thrombosis - DVT
Estimated Blood Loss - EBL
Emergency Department - ED
Hospital Day - HOD
Inferior Vena Cava - IVC
Jackson-Pratt - JP
Laboratory Risk Indicator for Necrotizing Fasciitis - LRINEC
Necrotizing Soft Tissue Infection - NSTI
Postoperative day - POD
Pulmonary Embolism - PE
Surgical Intensive Care Unit - SICU
Upper Respiratory Infections - URI

Patient Privacy
This report does not contain any personal information that could lead to the identification of the patient. All mentions of the patient were done so with the use of a pseudonym, or not at all.

Informed Consent
Informed consent was obtained via two physician phone consent with the patient.

Authorship
All authors attest that they meet the current criteria for authorship. All authors contributed equally to the writing of the report.

Conflict of Interest
All authors are without financial disclosures. No conflicts of interest declared by the authors.

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