Factitious Disorder Presenting with Attempted Simulation of Fournier's Gangrene

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ABSTRACT

Fournier's gangrene is a severe polymicrobial necrotizing fasciitis of the perineal, genital, or perianal regions. The classic presentation is severe pain and swelling with systemic signs. Crepitus and cutaneous necrosis are often seen. Characteristic CT findings include subcutaneous gas and inflammatory stranding. Unless treated aggressively, patients can rapidly become septic and die. Factitious Disorder is the falsification of one's own of medical or psychological signs and symptoms. Many deceptive methods have been described, from falsely reporting physical or psychological symptoms, to manipulating lab tests, or even injecting or ingesting foreign substances in order to induce illness. We present a case of a 35-year-old man with factitious disorder who attempted to simulate Fournier's gangrene by injecting his scrotum with air and fluid. We will review the clinical presentation and diagnosis of Factitious Disorder, as well as Fournier's gangrene.

CASE REPORT

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History:

A 35-year-old male with reported history of right orchiectomy two years ago for Non-Hodgkin's Lymphoma (NHL) presented to the emergency department (ED) with 3 days of testicular pain and scrotal swelling with fevers and chills. He reported pain at the orchiectomy site and in his remaining left testicle. As a possible inciting event, he had an ingrown hair on his scrotum a few days prior. He denied any urogenital or gastrointestinal symptoms.

The patient reported also having undergone chemotherapy and lymph node dissection for his NHL. His last treatment was two years prior, and his last surveillance imaging six months prior showed no disease. He denied any additional relevant medical or surgical history. His wife and daughter died in a car accident many years ago. He is on active military duty as a medic and lives on a base out of state. He denied drug or alcohol misuse.

Physical exam:

On physical exam, he was noted to be distressed. He was tachycardic at 122 beats per minute but mildly hypertensive at 156/100 mmHg. He was afebrile at 37.6˚C. His penis was normal. His right hemiscrotum was empty, and the scrotal skin was light pink with some induration, but no blanching erythema, crepitus or fluctuance. There were multiple scars on the scrotal skin. His tenderness to palpation was out of proportion to exam.

Laboratory studies:

Laboratory studies demonstrated a normal white blood cell count of 6.7 K/μL. His creatinine was 1.3 mg/dL (normal <1.2), but blood urea nitrogen was less than 5 mg/dL. Other laboratory values were within normal limits.

Imaging:

Testicular ultrasound demonstrated marked thickening of the scrotal skin without significant hyperemia. There were also shadowing echogenic foci within the deep subcutaneous tissues, concerning for gas-forming infection (figure 1). Contrast-enhanced CT of the abdomen and pelvis also demonstrated scrotal skin thickening, as well as gas within the inguinal canal and scrotum. The right testicle and spermatic cord were absent (figures 2 and 3). This was judged highly concerning for a necrotizing infection. There were little or no inflammatory changes in the pelvic or perineal fat, but this was not initially appreciated.

Clinical course:

He was seen by urologic surgery in the ED who administered broad-spectrum antibiotics and took him emergently to the OR for planned exploration and surgical debridement. Upon incising and entering the left scrotum, the surgeons were surprised to note that there was no evidence of any infection, necrotizing or otherwise. The wound was irrigated, cultures were taken, and the scrotum was closed. The urology team was understandably confused by the clinical situation, and suspected malingering or factitious disorder after some inconsistencies in his history came to light. They reached out to outside hospitals (OSH) to fact-check his medical history.

Records revealed that the patient presented 1 year ago to an OSH under very similar circumstances, with scrotal and retroperitoneal gas on imaging, and was treated for sepsis and Fournier's gangrene. Testicular exploration there was also negative for infection. He was admitted for one month, during which time he secretly performed venous and scrotal self-injections with contaminated water. He was bacteremic and fungemic. Blood cultures grew polymicrobial enteric organisms, with speciation suggesting feces or sewage water injection. He also had pulmonary nodules, biopsy-confirmed to be talc granulomas, consistent with intravenous drug use, which he denied. IV drug abuse was supported by the inability of his clinical team or peripherally inserted central catheter (PICC) nurses to gain IV access. At one point in his hospitalization he was observed conducting "suspicious activity," and was found with multiple paperclips, needles, syringes, and pills in his bed, which were confiscated. His history was also inconsistent. He told the physicians there that his orchiectomy was performed in Mexico for Fournier's gangrene contracted after scrotal surgery for penetrating trauma, not for lymphoma. At the end of the OSH admission a nurse caught him surreptitiously injecting his central line with a pink-colored fluid. Security was called and the patient was arrested because he had an outstanding warrant for burglary. He was discharged to jail.

Urology at our institution ordered a psychiatry consult. They noted that the patient was a very vague historian regarding many aspects of his life. His timeline and reported facts changed somewhat from telling to telling, including the year of death of his wife and daughter, his military service, occupation, and state of residence. He denied suicidality, or symptoms of depression, mania, or psychosis. He then admitted that he had presented to the OSH last year under false circumstances, but he affirmed that his concern for infection was genuine during this hospitalization. His blood cultures were negative. His wound culture was positive for a rare number of likely mixed skin flora. The diagnosis of factitious disorder was made, and the patient was discharged and scheduled for follow-up visit with his primary care provider.

DISCUSSION

Diagnosis and characteristics of Factitious Disorder

Formerly termed Munchausen's syndrome, but now called Factitious Disorder Imposed on Self (FD), is defined by the DSM V as the "deceptive falsification of medical or psychological signs and symptoms in oneself" [1]. In contrast, in Factitious Disorder Imposed on Another, formerly Munchausen's Syndrome by Proxy, an individual will falsify signs and symptoms in another individual. To meet criteria for FD, individuals must falsify disease, but not demonstrate pursuit of obvious external rewards. A variety of deceptive methods have been described in the literature, ranging from falsifying or exaggerating symptoms to ingesting foreign substances to manipulating laboratory test results. Previously reported cases of cellulitis from FD have involved air injections of the upper extremities, subfascial planes, and even the orbit [2-6]. A PubMed search using the terms "Fournier's gangrene factitious disorder" and "Fournier's gangrene Munchausen" yielded no other reports of simulated Fournier's gangrene (FG) secondary to FD.

The presentation of FD may closely resemble malingering, and it can be challenging for clinicians to be able to distinguish the two diagnoses. Both diagnoses involve the falsification of symptoms. However, in malingering, patients are doing so for some form of personal gain (money, avoiding legal consequences for criminal behavior, free meals). Our patient was not seeking secondary gain such money, time off work, or anything that would be suspicious for malingering. He also did not report any desire to make his testicle larger after orchiectomy, a potential cosmetic gain. Interestingly, his previous presentation at the OSH may have been malingering if he was attempting to avoid incarceration due to his outstanding arrest warrant at that time However, in the absence of any delusional or psychotic disorder, the patient fit all the criteria for diagnosis of FD at the time of presentation to our institution.

Some clinical and historical characteristics might alert physicians to suspect FD. For example, FD patients often seek treatment at multiple different hospitals. Their history can be inconsistent. Their clinical course may be atypical and not follow the natural history of the suspected disease. Other clues that may suggest FD are an unusually high number of hospital visits, symptom magnitude exceeding objective signs, physical findings of self-harm, and discovery of concealed instruments used to induce illness (e.g. syringes, needles) [7]. While

healthcare professionals may suspect FD in patients who present with symptoms that do not align with any defined medical diagnosis, it is important to note that FD can occur in patients with preexisting medical conditions as well. For example, patients with preexisting diabetes may surreptitiously increase their insulin dosage to produce physical symptoms from hypoglycemia. Often, FD patients have a medical background, and our patient fits that profile as he was a medic in the Armed Forces.

Etiology of subcutaneous gas

The etiology of scrotal gas can be classified into two categories based on the source of gas: internal or external. In cases where gas originated from an internal source, infection (cellulitis or FG), pneumothorax (spontaneous or traumatic), and perforated viscus should be considered [8-11]. Gas can migrate from the abdomen through a patent processus vaginalis, from the retroperitoneum through the inguinal canal, or from direct extension [11]. In cases where gas originates from an external source, trauma and iatrogenic are possible causes [12,13]. Reported iatrogenic causes include retroperitoneal surgery, chest drain insertion, and even endoscopic retrograde cholangiopancreatography (ERCP) [14-16].

Fournier's Gangrene

Pathology and Epidemiology

Fournier's gangrene is a severe necrotizing fasciitis of the perineal, genital, or perianal regions due to a mixed aerobic and anaerobic infection. The most common culprit organisms include E. coli, Klebsiella, Proteus, Staphylococcus, and Streptococcus [17]. When first described, FG specifically referred to an idiopathic infection, but the definition has expanded to include infections of other origins as well. It usually starts as a cellulitis, which leads to an endarteritis, and then a necrotizing gas-forming infection. Most commonly, the source of infections is the gastrointestinal tract (30—50%), especially perianal fistula or abscess. Less common etiologies are the genitourinary tract (20—40%), and skin (20%) [18,19]. Sometimes the cause is never identified.

FG is rare, and disproportionally affects males. Less than 0.02% of hospital admissions are for FG, and the overall incidence is 1.6/100,000 in males, with a predilection for those in the 50-79 year old age range, often diabetics or alcoholics. Twenty-70% of patients with FG have diabetes mellitus and 25-50% have a history of chronic alcoholism [18-20]. It can be quite lethal, even with adequate treatment, with a mortality rate of 15-50% [18-23].

Clinical Presentation And Diagnosis

Fournier's gangrene can present in a variety of ways, from appearing rapidly without any obvious precipitating factor, to being a slow and insidious infection. It is characterized by severe pain and swelling in the perineum and perianal region that can migrate along the anterior abdominal wall and into the gluteal, scrotal, and penile regions. Other common features are edema around the infection site, crepitus, and subcutaneous gas, with systemic signs including fever, tachycardia, and hypotension. Leukocytosis is common. As the subcutaneous infection and inflammation progress, visible necrosis may appear [17-19].

Radiologic studies are important for the identification of subcutaneous gas and provide useful information regarding the extent of disease and involvement of surrounding structures. However, imaging studies should not delay surgical therapy when there is clinical evidence of progressive soft tissue infection.

CT Imaging Findings

While FG is most commonly a clinical diagnosis, the classic finding of subcutaneous emphysema is best demonstrated by imaging studies, especially CT. CT can provide valuable information when the diagnosis or extent of disease are in question, as it has greater specificity than radiography, ultrasonography, or physical exam [24]. The characteristic finding is subcutaneous emphysema that dissects along fascial planes, which can extend from the perineal and perianal regions into surrounding structures, like the inguinal regions, thighs, abdominal wall, and retroperitoneum. Importantly, although subcutaneous emphysema is characteristic of FG, up to 10% of patients do not demonstrate this finding [24], so the absence of gas does not exclude it. However, the ability of CT to evaluate both superficial and deep fascial layers allows not only for recognition of FG, but also distinction from other entities like soft tissue edema or cellulitis. CT also provides great utility in assessing the etiology of gangrenous infection, as well as the presence of any fluid or abscess collections [24]. CT can also diagnose alternative causes of deep pelvic pain, including cystitis, prostatitis, proctitis, or perianal abscess, In addition, CT can be utilized in the post treatment period to monitor for resolution and improvement.

Other Imaging Modalities

Ultrasound is frequently the first test ordered in cases of suspected scrotal pathology, often in order to rule out torsion or epididymoorchitis. Ultrasound in patients with FG may demonstrate a thickened and edematous scrotal wall, usually with "dirty" shadowing (reverberation artifacts) reflecting foci of gas. There may be hydroceles or fluid collections. Scrotal contents can also be readily evaluated. The testicles and epididymes are usually normal with preserved color Doppler flow. Ultrasound is superior to radiography in demonstrating soft-tissue gas with a reported sensitivity up to 100% [25].

Radiography is not often part of the workup of suspected FG, but may be obtained if there is a significant component of abdominal pain. X-rays may reveal hyperlucencies representing soft tissue gas in the scrotal and perineal areas, sometimes extending into surrounding regions. Often gas is detected on radiographs before it is palpable on physical exam. Scrotal skin edema may also be observed. A significant weakness of radiography is limited sensitivity in detecting gas in deeper fascial layers. [26].

Magnetic resonance imaging (MRI) can also demonstrate characteristic findings. Characteristically, T1-weighted (T1W), dynamic contrast-enhanced (DCE) gradient echo (GRE) images demonstrate skin thickening and hyper-enhancement.

Fat suppressed T2-weighted (T2W) fast spin echo images (FSE) can demonstrate high signal intensity fluid and edema within the deep and superficial fascia. Fluid collections can be observed as low signal intensity on T1W images, with rim enhancement in the case of abscess. They are usually high signal intensity on T2W images [27]. Subcutaneous gas is manifest by very dark signal on both T1W and T2W weighted sequences, with susceptibility artifact on GRE. Because CT is quicker and more widely available, and because it is the most specific modality for evaluating FG [17], MRI is not commonly used.

Differential diagnosis and imaging characteristics

The differential for an acute non-traumatic scrotum includes: Fournier's gangrene, cellulitis, bowel-containing inguinal hernia, testicular torsion, and epididymo-orchitis.

Cellulitis is a skin infection that arises as a result of entry of bacteria through breaches in skin integrity. In contrast to FG, cellulitis involves the deep dermis and subcutaneous fat, but not deeper fascial layers. Clinically, the presentation of cellulitis is much less remarkable, presenting with swelling and redness. Systemic toxicity, rapid progression and tissue crepitus are not characteristic features, and their presence should alert the clinician to the presence of necrotizing fasciitis. In cellulitis, CT and ultrasound should not demonstrate subcutaneous gas or involvement of deep structures of the pelvis. However, the only way to reliably distinguish the two is by surgical exploration and visualization of the deep fascia.

In the setting of a bowel-containing inguinal hernia, US may demonstrate fluid- or air-filled loop of bowel in the scrotum. The presence of real-time peristalsis is diagnostic [28]. Radiography may demonstrate gas within bowel loops projecting over and below the inferior pubic ramus. Similarly, CT may demonstrate the presence of bowel loops that course through the inguinal canal [29]. Finally, MRI will demonstrate abnormal ballooning of the anteroposterior diameter of the inguinal canal and/or simultaneous protrusion of bowel within the inguinal canal [30].

In the setting of testicular torsion, Doppler US will demonstrate high resistance or absent blood flow in the testis. If ischemic, the testicle may be heterogeneous, enlarged, and hypoechoic [28]. Radiography and CT are not commonly used modalities in the evaluation of suspected torsion. However, MRI has been utilized and findings include decreased or absent perfusion in T1W GRE DCE-MRI, and a spotty or streaky pattern of low signal intensity in T2W images [31].

In the setting of epididymo-orchitis, US will demonstrate an enlarged epididymis, often hypoechoic relative to the testicle as a result of edema. The testicle may demonstrate heterogeneous echogenicity. Doppler US may demonstrate presence of hyperemia in both the testicle and the epididymis [28,32]. Ultrasound can also demonstrate the presence or absence of scrotal abscess. Radiography and CT are not commonly used modalities in the evaluation of suspected epididymo-orchitis. MRI may demonstrate an enlarged epididymis with possible decreased T2 signal, as well as hyper enhancement. Orchitis can manifest as areas of low signal intensity on T2W images. T1W DCE images may demonstrate heterogeneous hyperenhancement [33].

Management of Fournier's gangrene

After urgent resuscitation with fluids, immediate broad-spectrum antibiotic treatment with surgical debridement is required. Prompt and aggressive surgical debridement has been the most successful treatment approach for FG, as it is necessary to remove all non-viable tissue, halt progression of infection, and alleviate systemic toxicity [18]. Conservative treatment with antibiotics alone has not been successful at treating FG [34]. Patients often undergo an average of 3.5 procedures and may require cystostomy, colostomy, or orchiectomy [35]. Though the testes are classically spared in FG, up to 21% of patients with FG undergo orchiectomy due to non-viable testes [36]. Even with treatment, FG can be lethal, and has a mortality rate of 15-50% [18-23]

Conclusion

Although our patient did well postoperatively and experienced no complications, factitious disorder has the potential to lead to iatrogenic harm. Unfortunately, it is difficult to diagnose prospectively. Clues that may have informed the clinical team were the lack of elevated white blood cell count and fever, pain out of proportion to examination, the lack of severe perineal inflammation, as well as the presence of multiple unexplained scrotal scars. The radiologist interpreting the ultrasound correctly diagnosed scrotal gas and appropriately recommended a CT scan. Given the clinical concern and the alarming gas on CT, it was prudent of the radiologist to confirm the diagnosis of FG. Even in retrospect, noting the absence of significant inflammatory stranding and fluid, the diagnosis of FG would have been difficult to exclude. Ultimately, it is a clinical diagnosis, and the clinical presentation, as well as the imaging findings, were suspicious enough to prompt emergency surgery.

TEACHING POINT

Factitious disorder is the deceptive falsification of medical or psychological signs and symptoms in oneself and can be differentiated from malingering by the absence of pursuit of material gain. Fournier's gangrene is a necrotizing fasciitis involving the perineum. The physical exam findings are often striking, including edema, crepitus, and findings of sepsis. Imaging, especially CT, plays a key role in the diagnosis, demonstrating gas and inflammation in the deep tissues. Urgent surgery is usually needed.

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FIGURES

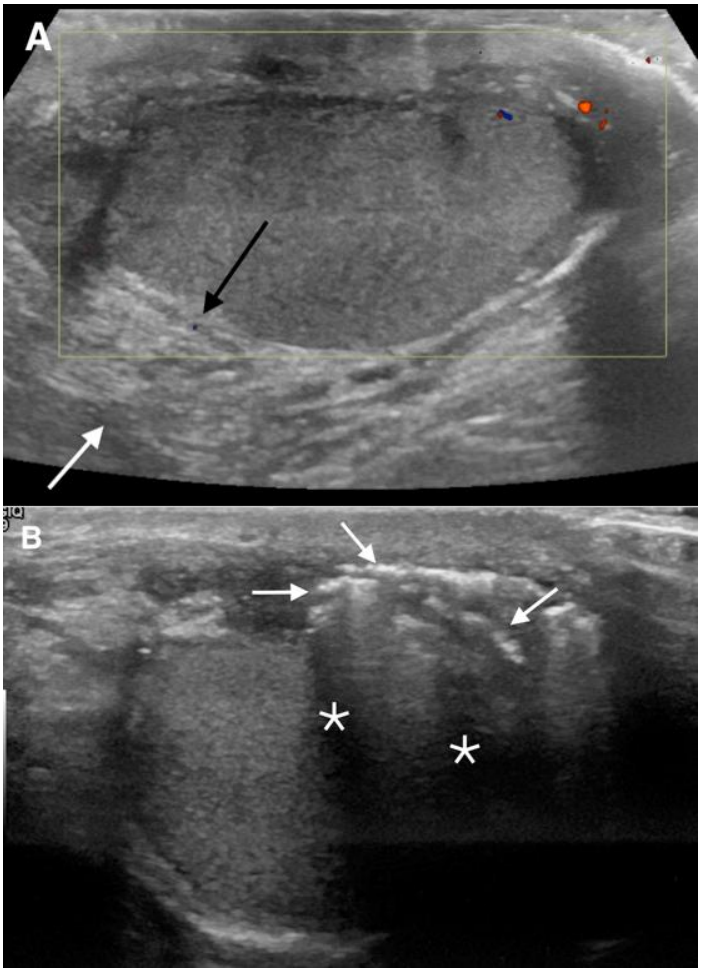

Figure 1: 35-year-old man with left scrotal pain and swelling. FINDINGS: Longitudinal color Doppler ultrasound image of the left testicle (A) demonstrates severe scrotal edema, measuring approximately 1.5 cm in thickness (between the black and white arrows). Note the lack of significant hyperemia. Longitudinal grayscale ultrasound image of the left testicle demonstrates multiple echogenic foci (arrows) representing gas, with posterior acoustic "dirty shadowing" (asterisks).
TECHNIQUE: Ultrasound performed on GE LOGIC E9 with ML 6-15 transducer at a frequency of 15.0 MHz

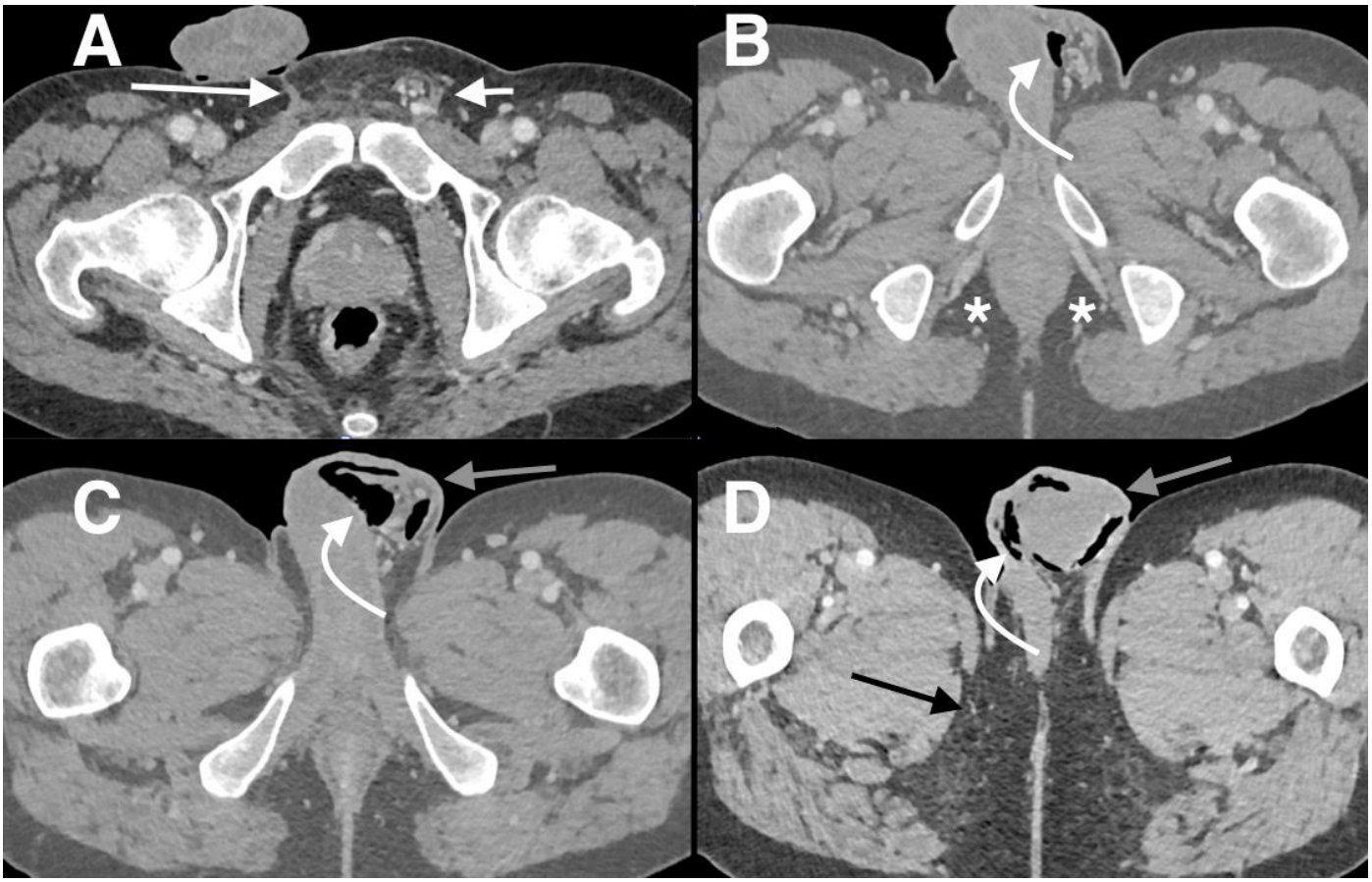

Figure 2: 35-year-old man with left scrotal pain and swelling.
Findings: Serial axial contrast-enhanced CT images of the pelvis from superior to inferior.
(A) There are postsurgical changes in the right inguinal canal (long arrow) with a normal appearing spermatic cord on the left (short arrow). Note the lack of extraperitoneal inflammation or gas surrounding the prostate and rectum.
(B) There is gas (curved arrow) in the left inguinal canal. Note the lack of inflammation in bilateral ischioanal fossae (asterisks).
(C) At the level of the penis there is gas (curved arrow) entering the scrotum. Scrotal skin thickening (gray arrow) is also noted.
(D) Gas (curved arrow) is seen surrounding left testicle. Scrotal skin thickening (gray arrow) is also noted. The right testicle is absent. Note the lack of inflammation in the perineum (black arrow).
Technique: Scan acquired on a Siemens Definition Flash dual source 2 x 128 detector CT in the portal venous phase. Slice thickness 1.0 cm. KVp 100, mA 451, 115 cc isovue 370 IV contrast injected at 2.9 mL per second.

Figure 3: 35-year-old man with left scrotal pain and swelling.
Findings: Coronal (A) and sagittal (B) contrast-enhanced CT images of the pelvis.
(A) There are postsurgical changes in the right inguinal canal (long arrow) with a normal appearing spermatic cord on the left (short arrow). There is gas (curved arrows) extending down the inguinal canal and surrounding the left testicle.
(B) There is gas (curved arrow) surrounding the left testicle. Note the paucity of inflammation in the perineum (black arrow).
Scan acquired on a Siemens Definition Flash dual source 2 x 128 detector CT in the portal venous phase. Slice thickness 1.0 cm. KVp 100, mA 451, 115 cc isovue 370 IV contrast injected at 2.9 mL per second.

Etiology	Infection, iatrogenic, perforated viscus, trauma,
Incidence	Less than 0.02% of hospital admissions. 1.6/100,000 in males
Gender ratio	Predominantly affects males
Age predilection	50-79 years of age
Risk factors	Diabetes and chronic alcoholism
Treatment	Surgical debridement
Prognosis	Guarded, 15-50% mortality rate
Findings on imaging	Subcutaneous emphysema, inflammation

Table 1: Summary table for Fournier's gangrene.

	Ultrasound	Radiography	CT	MRI
Fournier's gangrene	Thickened and edematous scrotal wall with "dirty" shadowing representing gas.	Hyperlucencies indicating soft tissue gas.	Subcutaneous gas along fascial planes, asymmetric deep fascial thickening, fluid and abscess	Gas in subcutaneous tissue, thickening of scrotal skin, and fluid collections. T1W GRE: heterogeneous enhancement. T2W FSE: high signal intensity in the deep and superficial fascial layers of the perineum
Indirect inguinal hernia	Fluid- or air-filled loop of bowel in scrotum. Presence of real-time peristalsis.	Air-filled bowel loops projecting over and below inferior pubic ramus	Hernia sac exists superolateral to inferior epigastric vessels and courses lateral to medial through the inguinal canal into the scrotum.	Abnormal ballooning of the anteroposterior diameter of the inguinal canal and/or simultaneous protrusion of fat and/or bowel within the inguinal canal
Testicular torsion	Absence of blood flow in testis. If ischemic, the testicle may be heterogeneous, enlarged, and hypoechoic.	Not commonly used	Not commonly used	Decreased or no perfusion in DCE-MRI and a spotty and/or streaky pattern of low or very low signal intensity in T2- and T2*-W images
Epididymo-orchitis	Inflamed and enlarged epididymis, often hypoechoic relative to testicle as a result of edema. Doppler US may demonstrate hyperemia.	Not commonly used	Not commonly used	Heterogeneous areas of low signal intensity on T2W images. Hypointense bands with heterogeneous enhancement on T1 W GRE

Table 2: Differential table for Fournier's gangrene.

ABBREVIATIONS

CT = computed tomography
DCE = dynamic contrast-enhanced
DSM = Diagnostic and Statistical Manual of Mental Disorders
ED = emergency department
ERCP = endoscopic retrograde cholangiopancreatography
FD = factitious disorder
FG = Fournier's gangrene
FSE = fast spin echo
GRE = gradient echo
IV = intravenous
MRI = magnetic resonance imaging
NHL = Non-Hodgkin's Lymphoma
OSH = outside hospital
PICC = peripherally inserted central catheter
T1W = T1-weighted
T2W = T2-weighted
US = ultrasound

KEYWORDS

Munchausen's syndrome; Factitious disorder; Fournier's gangrene; simulated Fournier's gangrene; scrotum; subcutaneous emphysema; ultrasound; CT