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Urethral fistula as an Unusual Complication of Fournier's Gangrene: a Case Report and Literature Review

Chih-Weim Hsiang[1], Guo-Shu Huang[1], Wen-Cheng Liu[2], Hsian-He Hsu[1], Seng-Tang Wu[3], and Wei-Chou Chang[1*]

[1]Department of Radiology; [2]Department of Internal Medicine; [3]Division of Urological Surgery, Department of Surgery, Tri-Service General Hospital, National Defense Medical Center, Taipei, Taiwan, Republic of China

Urethral fistula is a very rare complication of Fournier's gangrene. A persistently discharging penile sinus should prompt physicians to consider this unusual entity. To our knowledge, only two similar cases have been reported in the literature. Preoperative multidetector computed tomography (MDCT) images were not used to show the route of fistula in either of them, but the route was discovered during surgery. Here, we report a rare case of a 43-year-old, otherwise healthy, man who developed a ventral side urethral fistula as a result of Fournier's gangrene found in preoperative images. Reconstructed images proved to be a valuable tool to highlight the path and extent of this unusual complication of Fournier's gangrene before surgery.

Key words: Fournier's gangrene, urethra, fistula, multidetector computed tomography (MDCT), penis, scrotum, corpora cavernosum

INTRODUCTION

Fournier's gangrene represents a rare urologic emergency with a high mortality rate. Diagnosis of the gangrene is primarily based on clinical presentation. Although initially thought to be an idiopathic entity, Fournier's gangrene has been shown to present in the vast majority of cases, as either perineal or genital skin infections.[1] Lack of awareness of the progression and extent of the disease can result in rapid multiple organ failure and death. Because of the fulminating process and potential complications of the disease, preoperative CT can be an important tool with which to diagnose its extent as early as possible. Our case presents a urethral fistula detected by reconstructive CT before surgery. This is the third case reported in the literature.

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*Corresponding author: Wei-Chou Chang, Department of Radiology, Tri-Service General Hospital, National Defense Medical Center; No. 325, Sec. 2, Cheng-gong Road, Taipei 114, Taiwan, Republic of China. Tel: +886-2-87927244; Fax: +886-2-87927245; E-mail: weichou.chang@gmail.com

CASE REPORT

A 43-year-old man presented to our emergency department with progressive lower abdominal pain, groin pain, dysuria, and a foul-smelling penile discharge for 3 days. He had no history of diabetes mellitus or other medical comorbidities. He denied heavy alcohol use.

On measurement of vital signs, he had tachycardia, with a pulse rate of 110 beats/min, blood pressure of 95/70 mmHg, and was febrile with a body temperature of 38.6 ^{o}C. Physical examination revealed erythema, swelling and tenderness over the penis and scrotum. Gangrenous skin with a suspected sinus of the ventral penis was also noted. The erythema extended to the inguinoscrotal region and lower abdomen with tenderness. The remainder of his abdomen was generally soft and nontender. No perianal lesion was seen. Laboratory examination revealed leukocytosis with a white blood cell count of 13200/mm^3, C-reactive protein of 32.87 mg/dl, blood urea nitrogen of 27 mg/dl, and serum creatinine of 1.2 mg/dl. Urine analysis disclosed a red blood cell count of 35 per high power field, numerous white blood cells, and bacteriuria. Based on the clinical course and laboratory data, urinary tract infection with sepsis was suspected.

An abdominal radiograph showed soft-tissue opacity in the rectum, causing diffuse gas-filled distension of the proximal colonic bowel loops. There was no evidence of extraluminal free air accumulation or pneumatosis

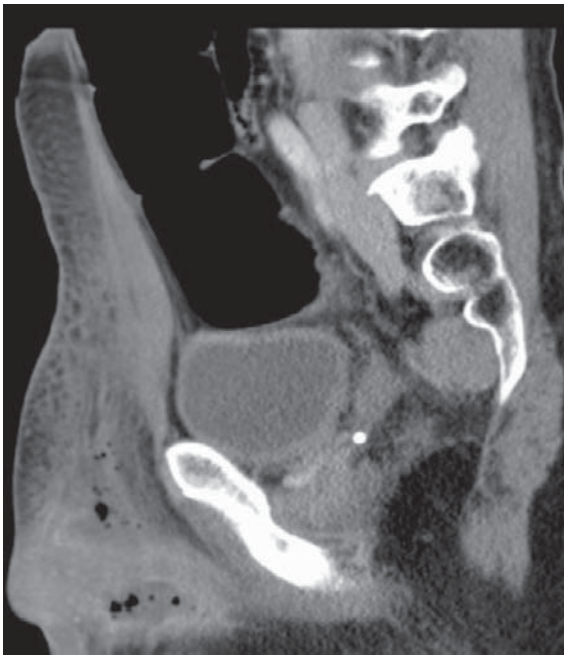

Fig. 1 An oblique sagittal MDCT image showed markedly fat stranding, subcutaneous emphysema, fluid accumulation and abscess formation extending to the perineum and abdominal wall.

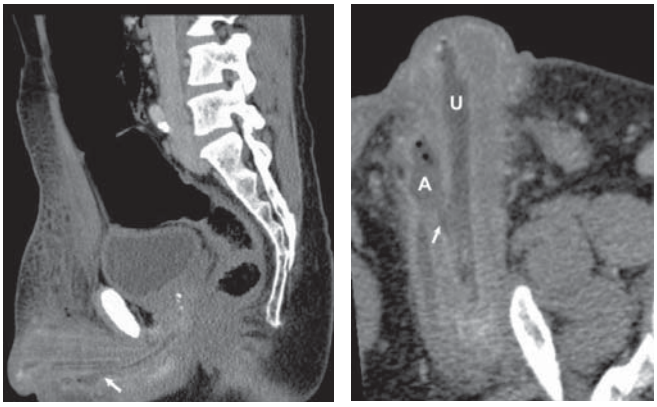

Fig. 2 (a) An oblique sagittal MDCT image showed a fistula (arrow) arising from the ventral side of the bulbous urethra and extending to the adjacent skin was evident on reformatted oblique sagittal image.
(b) An oblique axial MDCT image showed abscess-like fluid with tiny gas pockets tracked in the right corpora cavernosa and formed a fistula to the bulbous urethra (arrow). (U: urethra; and A: abscess)

intestinalis over this region. To further characterize the disease entity, MDCT was performed and revealed a markedly enlarged and thick-walled scrotal sac containing fluid and foci of gas that extended cranially to the right inguinal canal, perineum, and the lower abdominal wall. Subcutaneous fatty stranding with emphysematous change was seen around the involved structures (Figure 1). Free gas in the urinary bladder was found. Fluid and gas pockets tracking in the right corpus cavernosum and a fistula formation on the ventral side of bulbous urethra was also identified in the multiplanar reformatted images (Figure 2). The images confirmed the diagnosis of Fournier's gangrene, complicated by a urethral fistula.

The patient was taken to the operating room for urgent surgical management. Debridement of the scrotum and anterior abdominal wall and drainage of the corpus cavernosum abscess was performed. A defect at the ventral side of the penis forming an urethrocutaneous fistula with the bulbous urethra was seen (Figure 3). A urinary diversion of suprapubic cystostomy was performed to allow voiding.

The patient received a long course of empirical antibiotics with intravenous piperacillin/tazobactam 4.5 g every 6 h and six aggressive surgical debridements of all necrotic tissue. Wound culture disclosed polymicrobial infection with *Viridans streptococcus*, *Escherichia coli*, and *Bacteroides fragilis*, which were all susceptible to piperacillin/tazobactam. Two months after admission, he was discharged in stable condition with a suprapubic cystostomy. Six months later, he underwent urethral reconstruction with a thick-split skin graft taken from his left thigh while the infection had been controlled well.

DISCUSSION

Fournier's gangrene is a life-threatening urological emergency of gangrenous infection in the perineal, genital, and perianal region. It was first described by Alfred Jean Fournier in 1883 as necrotizing fasciitis involving the male genitalia and awareness of this disease has increased in recent years. The wide spectrum of its complications, including penile gangrene, colorectal infection, urethral stricture, urethral fistula, fatal tetanus, squamous cell carcinoma, ketoacidosis, sepsis, and death, have been noted.1 Some of these complications are serious and require repeated surgical treatment and cosmetic reconstruction.

Urethral fistula is one of these serious complications and is extremely rare. Only two cases have been reported.[2-3] They have variable presentations and are difficult to identify clinically. In 2004, Grandhi et al. published the first description of a case of Fournier's

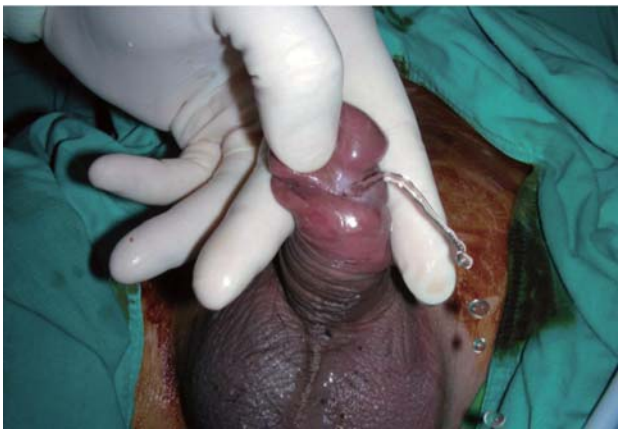

Fig. 3 During intraoperative normal saline injection test, the picture showed water leakage from the urethral fistula

gangrene complicated by a urethral fistula. A patient with a case of diabetes mellitus presented with penile swelling and redness. Urine leaking from the penis was not found initially, but was found after surgical debridement. A urethral fistula, located on the dorsal aspect of the penis, was identified after repeated surgery. Lee et al. published the second description of a case in 2011. A heavy drinker presented with scrotal swelling and urine leakage through his scrotal wall. A urethral fistula, located on the ventral side of the penis, was identified during exploratory surgery. To our knowledge, our case is the third reported in the literature. The patient had no comorbidity and no associated clinical presentation.

The exact pathological mechanism of the urethral fistula is unknown. We reviewed the three cases and propose a mechanism based on our conclusions. Perineal infection in Fournier's gangrene involves the periurethral gland or a pre-existing urethral diverticulum, which leads to periurethral abscess formation. Then, subsequent rupture of this abscess can result in the urethral fistula.[2-3]

It is crucial to recognize Fournier's gangrene because it has a high mortality rate, ranging from 15% to 50%.[4] Clinically, the patient presents with scrotal swelling, pain, hyperemia, pruritus, a foul-smelling discharge, and fever as common symptoms.[5] Diabetes mellitus, older age, chronic ethanol abuse, and other debilitating states have been found as risk factors for Fournier's gangrene.[6] Early diagnosis, immediate surgical debridement, and aggressive antibiotic treatment are indicated.

MDCT is the only investigative modality to confirm the diagnosis of Fournier's gangrene and its etiology for appropriate surgical treatment.[5] Subcutaneous emphysema, which has been reported in up to 90% of patients, is a hallmark of Fournier's gangrene.[7] Furthermore, multiplanar reformatted imaging is useful in assessing the extent of the disease and its complications. In our case, reformatted oblique sagittal images revealed the path of the urethral fistula as a diagnostic image and altered the initial approach to surgical planning with the creation of a urinary diversion.

The treatment strategy for urethral fistula must be individualized according to its size, location, and the surrounding tissue loss. MDCT and multiplanar reformatted imaging all offer relevant information before surgery. The three cases were all managed by urinary diversion and delayed urethroplasty until infection had been controlled. Lee and Hong used a suprapubic catheter because of bulbous urethral fistula necrosis associated with possibly preceding urethral stricture and periurethral abscess, as found in our case. To recover voiding function, adequate debridement of necrotic tissue, infection control, and then closure of the fistula by uroplasty with a skin graft are considered in the subsequent treatment.[2]

In conclusion, urethral fistula as a complication of Fournier's gangrene is a rare entity. We believe that MDCT and reconstructive CT offer valuable information with which to evaluate the Fournier's gangrene and its associated complications before surgery.

DISCLOSURE

All authors declare no competing financial interests.

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