CASE REPORT

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Acute peritonitis secondary to postpartum gangrenous cystitis

Peritonitis aguda secundaria a cistitis gangrenosa posparto

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ABSTRACT
Gangrenous cystitis is an extremely rare infectious disease. Its etiology is multifactorial and the main precipitating factor is overdistention of the bladder due to obstruction. Bladder involvement varies from mucosal and submucosal necrosis to necrosis of the entire wall, which can lead to acute peritonitis. Diagnosis is difficult due to its low incidence and nonspecific symptomatology, similar to that of other urinary infectious conditions, resulting in late diagnosis and increased morbidity and mortality. Early and aggressive surgical management is essential to reduce frequency of complications associated with this condition. A case of acute peritonitis secondary to postpartum gangrenous cystitis is presented.
Key words: Cystitis, Gangrene, Peritonitis.

RESUMEN
La cistitis gangrenosa es una enfermedad infecciosa extremadamente rara. Su etiología es multifactorial y el principal factor precipitante es la sobredistensión de la vejiga debida a obstrucción. La afección vesical varía desde necrosis de mucosa y submucosa hasta necrosis de toda la pared, que puede ocasionar peritonitis aguda. El diagnóstico es difícil debido a su baja incidencia y a la sintomatología inespecífica, similar a la de otras afecciones infecciosas urinarias. Ello resulta en un diagnóstico tardío y mayor morbimortalidad. El manejo quirúrgico temprano y agresivo es fundamental para disminuir la frecuencia de complicaciones asociadas a esta condición. Se presenta un caso de peritonitis aguda secundaria a cistitis gangrenosa posparto.
Palabras clave. Cistitis, Gangrena, Peritonitis.

Introduction

Gangrenous cystitis is an extremely rare infectious condition, with only 33 cases reported in the last 75 years, but potentially fatal[1]. Its severity varies from mucosal-submucosal necrosis to necrosis of the entire bladder wall[2,3]. The etiology is multifactorial and symptoms are nonspecific, which makes it indistinguishable from other common urological conditions, so the precise diagnosis can be difficult and is generally performed when there are manifestations of acute peritonitis. This is aggravated by its very low incidence, which contributes to high morbidity and mortality[1,4]. A case of acute peritonitis secondary to postpartum gangrenous cystitis is presented.

Clinical case

The patient was 26 years old, gestation 1, para 1, who attended the emergency room because of moderate to severe abdominal pain without heartburn, accompanied by urinary retention, decreased micturition frequency, fever, nausea and vomiting of 4 days of evolution. She reported an instrumental vaginal delivery 8 days earlier and during pregnancy she had been diagnosed with pyelonephritis, for which she was hospitalized and received intravenous antibiotic therapy. The patient denied a history of voiding dysfunction and other important personal and/or family medical history.

Physical examination showed that she was feverish (39.9°C), hypotensive (90/50 mm Hg), tachycardic (140 beats / minute) and tachypneic (22 breaths / minute). The abdomen was slightly distended, but very painful to shallow and deep palpation, accompanied by generalized rigidity. The hydroaerial noises were diminished. Gynecological examination showed a normal cervix, with sero-bloody lochia and a uterus with normal involution. No abnormalities were detected on speculoscopy or rectal examination. Bladder drainage showed urinary retention (more than 2 liters of urine), with slightly hematuric urine.

Laboratory tests results were: leukocytes 16,700 cells / mL with a predominance of neutrophils (89%); C-reactive protein 19 mg / dL and serum creatinine 1.5 mg / dL. Rest of the kidney and liver function tests, coagulation profile, and electrolytes were within normal limits. Urinalysis indicated uncountable number of leukocytes and pyuria (15-20 cells per field). Plain abdominal radiography showed multiple dilated small bowel loops filled with fluid, with no evidence of subdiaphragmatic free air. On ultrasound, the kidneys appeared normal and unobstructed, while the bladder was distended, almost reaching the level of the umbilicus, with no filling defects in the lumen. The uterus and adnexa were within normal limits. In view of the findings, the patient was hospitalized and treated with analgesics, empirical intravenous antibiotic therapy, and fluid replacement.

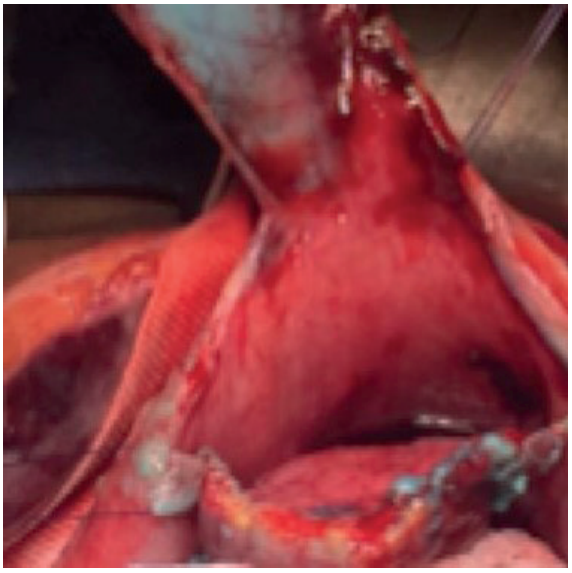
The following day, the patient's clinical status deteriorated. She remained feverish, oliguric, presenting increased abdominal distention, absence of bowel sounds and mental confusion. Abdominal computed tomography images showed abundant amounts of free fluid in the cavity, with moderate bilateral dilatation of ureters and pyelocaliceal system, gas within the anterior wall of the bladder and dilated intestinal loops, without apparent obstruction of the passage of intestinal contents. Instillation of contrast through the catheter into the bladder revealed extravasation into the peritoneal cavity. In view of the findings, the diagnosis of bladder perforation was made and it was decided to perform an exploratory laparotomy.

During surgery, signs of acute generalized peritonitis were found with approximately 5 liters of free purulent fluid in the abdominal cavity and distended bowel loops covered by serous material. There was evidence of necrosis of the perivesical fat with abundant purulent secretion in the retropubic space. The bladder was enlarged, thick walled, and the dome had two defects with greenish, necrotic borders separated by normal tissue, which were probably the cause of acute peritonitis. After incision of the bladder, full wall thickness necrosis was revealed, and the bladder mucosa had several areas of petechial hemorrhage and necrosis (Figure 1). Both ureteral orifices, as well as the trigone, were preserved, with no evidence of necrosis. Partial cystectomy was performed with debridement of the necrotic tissue and preservation of both trigone and ureters, closing in two layers with 2-0 vicryl. The rest of the abdominal cavity was explored, peritoneal lavage was performed and a suprapubic catheter and two surgical drains were placed.

Pathological evaluation revealed total necrosis of the bladder mucosa with partial necrosis of the muscular wall and focal infiltration of lymphocytes and plasmacytes. The results of the culture of the resected bladder tissue showed growth of *Escherichia coli*. The final diagnosis was gangrenous cystitis.

The patient was initially transferred to the intensive care unit for fluid replacement, ventilatory support and treatment with broad-spectrum antibiotics, where she remained for 5 days. The postoperative period passed without complications, with primary healing of the operative wound. She

Figure 1. Internal bladder mucosa during partial cystectomy showing petechial hemorrhages and necrotic areas.



was subsequently transferred to hospitalization and discharged 14 days after surgery, with urethral catheter and ambulatory cystography.

Discussion

Postpartum gangrenous cystitis is extremely rare in current clinical practice, with only a few cases described in the literature[5]. It was initially described in 1650. It usually appears in elderly patients and predominantly affects patients with associated comorbidities, such as diabetes, spinal cord injuries, pelvic neoplasms or lithiasis[3].

The etiology is multifactorial and the identification of a single origin is difficult[6]. The causes can be divided into indirect and direct. Indirect causes are those that interfere with blood flow, causing tissue deterioration and bladder necrosis. Pressure from the inside (overdistention of the bladder due to chronic urinary retention) or pressure from the outside (poor position of the pregnant uterus, prolonged labor, pelvic neoplasms and surgical interventions) are the main conditions associated with appearance of the clinical picture. Ligation, embolism or thrombophlebitis of the arterial and / or venous vessels can also cause this condition. On the other hand, direct factors, potentially through their toxic effects, can cause bladder wall cells death. These causes include intravesical agents, pelvic radiation and severe systemic infections. Gas-forming microorganisms have been implicated in cases of patients diagnosed with uncontrolled diabetes mellitus[7-11].

Although the exact etiology of gangrenous cystitis is unknown, alterations in bladder blood flow indirectly cause necrosis of the bladder wall. The resulting ischemia varies in severity, from mucosal and submucosal necrosis to full-thickness necrosis of the bladder wall[12,13]. Cases of postpartum gangrenous cystitis are associated with prolonged labor and instrumental delivery that produce alterations in the transmission of electrical impulses of the pelvic nerves that cause bladder hypotonia, excessive distention and gangrenous cystitis[4]. On rare occasions, gangrenous cystitis can lead to the formation of colovesical fistulas[14].

Diagnosis of gangrenous cystitis is usually difficult, since the initial symptomatology (abdominal pain, microscopic / macroscopic hematuria or pyuria) is nonspecific and similar to other conditions, such as cystitis. The urine is foul-smelling and contains pus along with exfoliated debris and thus causes retention. As the disease progresses, abdominal manifestations predominate with findings of acute peritonitis and deterioration of the general condition[5]. Peritoneal involvement presents pathognomonic signs of an acute abdomen: rigidity of the abdominal wall and absence of hydroaerial sounds[15].

Laboratory findings are nonspecific and report leukocytosis and high erythrocyte sedimentation rate. Urine cultures can identify the causative microorganisms and antibiotic susceptibility. Computed tomography, cystography and cystoscopy are useful to establish the precise diagnosis. Cystoscopy could also serve as a diagnostic tool, especially because it allows direct visualization and collection of specimens for biopsy and urine culture. Computed tomography, which is another useful diagnostic tool, is usually performed when patients present with acute abdominal symptoms[12].

In most cases, due to extensive bladder necrosis, emergency surgery is necessary. Conservative treatment (antibiotic therapy along with adequate bladder drainage and use of hyperbaric oxygen) can be performed in stable patients with mucosal involvement only. Surgical treatment should be early and aggressive and includes extensive debridement of necrotic portions of the bladder. In most cases, the trigone remains viable (this anatomic portion is additionally irrigated by arterial branches that irrigate the ureters) and, consequently, total cystectomy with urinary diversion is rarely necessary[7,8]. After resolution of the clinical picture, it is possible to attempt bladder regeneration[8,9,12].

However, despite adequate treatment, the mortality rate is approximately 35%. This is mainly due to diagnostic delay due to nonspecific symptomatology, mixed infections with resistant strains and associated comorbidities[7,13].

In conclusion, gangrenous cystitis is a rare but potentially severe condition. Early and timely diagnosis of acute peritonitis due to this condition is critical to improve prognosis. Early and aggressive intervention, together with adequate drainage and adequate antibiotic therapy, reduces the risk of associated morbidity and mortality.

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