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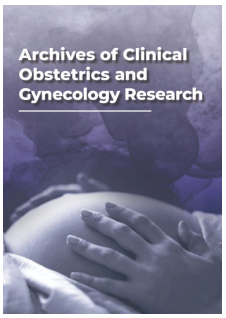
Case Report



Spontaneous Bladder Rupture Presenting as a Urinary-Cutaneous Fistula During Chemotherapy in a Radiated Pelvis

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Abstract

Spontaneous rupture of the urinary bladder is a rare event that may be caused by prior radiotherapy, bladder dysfunction, alcoholic intoxication, cancer invasion, and inflammation. We herein describe a 58-year-old woman diagnosed with spontaneous bladder rupture presenting with a urinary-cutaneous fistula during chemotherapy for peritoneal cancer. Twenty-five years ago, the patient underwent radical hysterectomy followed by radiotherapy for cervical cancer and developed neurogenic bladder. She was referred to our hospital due to pleural effusion and ascites. After exploratory laparotomy, she was diagnosed with stage IVB peritoneal cancer. She received chemotherapy with paclitaxel and carboplatin. While chemotherapy was effective, she was hospitalized for left pyelonephritis and septic shock. After receiving antibiotics, she was recommended for self-catheterization for urination. After the fifth course of chemotherapy, she presented with a large amount of fluid leaking from the lower part of her abdominal wound. Computed Tomography (CT) and cystography suggested that extraperitoneal rupture of the urinary bladder had led to an abscess of the space of Retzius, with urine and pus leaking from the urinary-cutaneous fistula. A urinary balloon catheter was placed and the deep abscess cavity was washed out. After 2.5 months, both the extraperitoneal bladder rupture and the fistula resolved with conservative management. Two years after resuming chemotherapy, the patient died from the progression of peritoneal cancer.

Keywords: Chemotherapy; Peritoneal cancer; Radiation; Spontaneous bladder rupture; Urinary-cutaneous fistula

Background

Spontaneous rupture of the bladder is a relatively rare condition associated with factors such as chronic bladder inflammation, malignant invasion, prior pelvic radiotherapy, neurogenic bladder, and alcoholic intoxication [1]. Cases have been reported following radiotherapy for cervical cancer [2-4]. Bladder rupture typically presents with acute abdominal symptoms, requiring medical attention. We herein report a case of spontaneous bladder rupture that occurred during chemotherapy for peritoneal cancer in a patient with a history of surgery and radiotherapy for cervical cancer. Rupture resulted in the formation of a urinary-cutaneous fistula through the surgical scar, which subsequently improved with conservative treatment.

Case report

The patient was a 58-year-old woman (gravida 1, para 1) with a previous medical history of cervical cancer treated 25 years earlier with radical hysterectomy followed by postoperative radiotherapy. She presented with progressive dyspnea and abdominal distension over a period of four months and was referred to our hospital due to the presence of pleural and peritoneal effusion. Cytological analyses of both pleural and ascitic fluid revealed class V adenocarcinoma. Positron Emission Tomography/Computed Tomography (PET-CT) and Magnetic Resonance Imaging (MRI) showed extensive peritoneal cancer involving the omentum, with no enlargement of the uterus or ovaries. She underwent exploratory laparotomy, during which a portion of the disseminated lesions was resected. A histopathological examination confirmed the diagnosis of high-grade serous carcinoma, and she was subsequently diagnosed with stage IVB peritoneal cancer. Chemotherapy with paclitaxel and carboplatin was initiated on postoperative day 18. Prior to the onset of peritoneal cancer, the patient had experienced diminished bladder sensation, and was known to have neurogenic bladder as a complication of prior radical hysterectomy. Although she was instructed by a urologist to perform clean intermittent self-catheterization,

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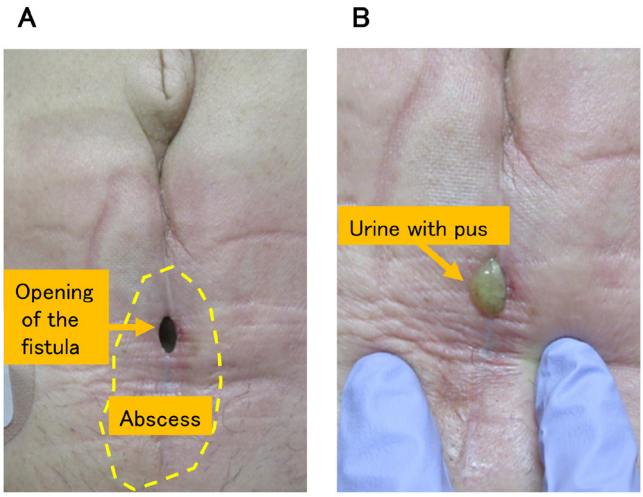

Figure 1. *An opening was observed at the lower part of the lower abdominal surgical scar (A), and compression caused the leakage of urine mixed with a purulent discharge (B).*

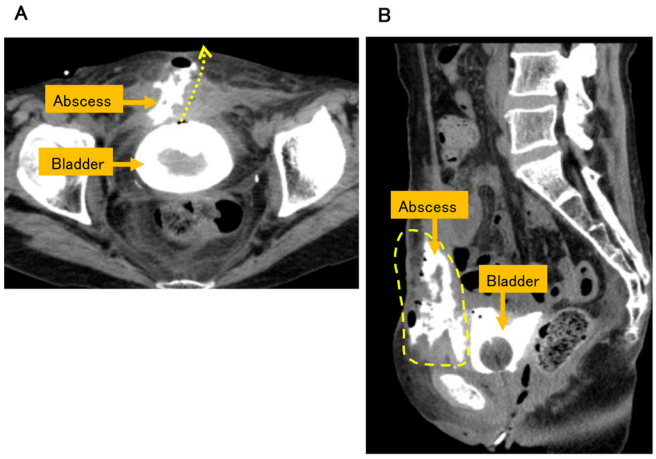

Figure 2. *CT image after cystography. Contrast medium was observed flowing into the abscess cavity in the space of Retzius. These findings were consistent with urine leakage from bladder perforation/rupture through the abscess cavity to the surgical wound.*

she was unable to comply due to fatigue associated with chemotherapy. During the fourth course of chemotherapy, she developed left-sided pyelonephritis, which progressed to septic shock on postoperative day 124. Her condition improved following antibiotic treatment and the placement of a urethral balloon catheter. After the balloon catheter was removed at discharge, she performed self-catheterization infrequently. The residual urine volume was not monitored, and the frequency of urination decreased. On day 13 of the fifth course of chemotherapy, she presented to the Emergency department with copious fluid discharge from the lower portion of a previous lower abdominal surgical scar. Urine mixed with purulent fluid was leaking from an opening at the lower part of the scar (Figure 1). Cystography followed by CT revealed extraperitoneal perforation/rupture of the bladder extending into the space of Retzius with the formation of an abscess and urine with purulent fluid leaking through the surgical scar, which resulted in the development of a urinary-cutaneous fistula (Figure 2). There was no evidence of direct tumor invasion into the bladder wall by peritoneal cancer. Although the external opening was approximately 1 cm in diameter, an internal abscess cavity had formed. Therefore, the wound was surgically opened along the previous incision for 4 cm, and the abscess cavity was irrigated repeatedly. Povidone-iodine ointment was applied, and conservative treatment was continued. A urethral balloon catheter was maintained throughout. After 2.5 months, the urinary-cutaneous fistula closed and chemotherapy was resumed. Two years later, the patient developed brain metastases and ultimately passed away due to disease progression from peritoneal cancer.

Discussion

Bladder rupture is most commonly caused by trauma, while spontaneous bladder rupture is relatively rare. Several factors have been associated with an increased risk of spontaneous rupture, such as a history of bladder surgery, bladder diverticula, bladder tumors, chronic bladder inflammation, cancer invasion, prior pelvic radiotherapy, neurogenic bladder, and alcoholic intoxication [1-6]. The present case had undergone radical hysterectomy for cervical cancer and had a history of both neurogenic bladder and pelvic radiotherapy, placing her at a high risk of bladder rupture.

There are two types of bladder rupture: intraperitoneal rupture, in which urine overflows into the peritoneal cavity, and extraperitoneal rupture, in which urine leaks outside the peritoneum, often resulting in a peri-vesical abscess [7]. Clinical symptoms may include abdominal pain, abdominal distension, urinary difficulty, oliguria or anuria, hematuria, fever, vomiting, and diarrhea [1]. Intraperitoneal rupture must be differentiated from gastrointestinal perforation or perforated peritonitis. In the present case, urinary difficulty due to neurogenic bladder, prior radiation fibrosis, and increased intra-abdominal pressure from chemotherapy may have contributed to bladder overdistension and straining during urination, resulting in a small extraperitoneal bladder rupture. Infected urine likely accumulated through the rupture site into the space of Retzius, forming an abscess. As the internal pressure of the abscess increased, the infection extended towards the vulnerable surgical scar, eventually leading to the leakage of urine through the skin. Prior to the episode of bladder rupture, the patient had developed pyelonephritis complicated by septic shock. Blood cultures at that time revealed Escherichia coli, which was also isolated from the urine. However, Klebsiella oxytoca was detected in both the abscess associated with bladder rupture and in the urine culture obtained during that period, suggesting that urinary bacteria were the source of the peri-vesical abscess. This bacterial shift was presumed to represent an evolving flora due to catheterization rather than a superinfection. Furthermore, cystography and CT revealed no significant evidence of peritoneal cancer invasion into the bladder wall, suggesting that bladder rupture due to direct tumor infiltration from outside the bladder was unlikely.

Cases in which spontaneous bladder rupture is followed by the formation of a urinary-cutaneous fistula are extremely rare. Raup et al. reported 21 cases of urinary-cutaneous fistulas in patients with neurogenic bladder, where the underlying conditions included traumatic spinal cord injury, cerebral palsy, transverse myelitis, and syringomyelia [8]. Johnsen et al. also presented six cases in which the conservative

management of traumatic extraperitoneal bladder ruptures led to the formation of urinary-cutaneous fistulas [9]. Although the present case did not involve traumatic rupture, an abscess formed as a result of extraperitoneal bladder rupture and eventually created a fistulous tract through the fragile surgical scar. Similar to our case, Toufique et al. reported a fistula from the bladder to the abdominal wall surgical scar following a cesarean hysterectomy [10]; however, fistula formation from the bladder to a cutaneous scar is extremely rare compared to those to the vagina [11].

Treatment strategies for bladder rupture include conservative management with urethral catheterization and surgical repair. In cases diagnosed with intraperitoneal rupture, emergency laparotomy is often required due to acute peritonitis. Zhang et al. reported 713 cases of spontaneous rupture of the urinary bladder [1]. While most intraperitoneal ruptures require surgical approaches, conservative management can be considered in cases of extraperitoneal rupture [1]. The choice between long-term urethral catheter placement and surgical bladder repair must be made on a case-by-case basis. In the present case, several factors supported the decision for conservative treatment: a history of pelvic radiotherapy, neurogenic bladder secondary to prior radical hysterectomy, and clear evidence of extraperitoneal rupture with the formation of a fistula from an abscess of the space of Retzius to a surgical scar. Therefore, conservative management was selected, and bladder rupture and the urinary-cutaneous fistula both resolved within 2.5 months. Although spontaneous bladder rupture is a rare condition, it is essential to consider its possibility in high-risk patients and select an appropriate treatment strategy based on the individual clinical context.

Conflict of Interest

The authors declared that they have no competing interests.

Grant

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