



## Case report

# Spontaneous bladder rupture attributable to a radical hysterectomy-associated neurogenic bladder in patients with cervical cancer: A case report and literature review

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## ABSTRACT

**Introduction and importance:** Spontaneous bladder rupture (SBR) is an extremely rare urological emergency. Herein we report a rare case of SBR in a postoperative cervical cancer patient, which was attributable to bladder distension due to a radical hysterectomy-associated neurogenic bladder.

**Case presentation:** A 74-year-old nulliparous Japanese patient with cervical cancer (pT1b3N0M0) presented with acute abdominal pain nine days after a radical hysterectomy. The pretreatment workup included *plain computed tomography (CT) revealed the presence of ascites in the absence of gastrointestinal perforation.* The patient was initially diagnosed with generalized bacterial peritonitis and treated with antibiotics. Urine outflow was noted 5 days later from the vaginal stump. Subsequent *contrast-enhanced CT demonstrated a bladder wall defect with presence of contrast medium in the abdominal cavity.* The patient was diagnosed with SBR and was conservatively treated with antibiotics and prolonged catheterization (4 weeks); these measures showed no signs of therapeutic efficacy. The patient was subsequently treated surgically with an ileal conduit urinary diversion. The patient is currently free of disease.

**Clinical discussion:** A literature review revealed that a history of pelvic radiotherapy is the main predisposing factor for SBR in women with cervical cancer. Our case serves to alert physicians that SBR should be considered a differential diagnosis in postoperative cervical cancer patients without a history of pelvic radiotherapy who experience generalized peritonitis symptoms or present as an acute abdomen.

**Conclusion:** SBR can develop in cervical cancer patients without a history of radiotherapy. This differential diagnosis should be considered in patients with a radical hysterectomy-associated neurogenic bladder.

## 1. Introduction

Spontaneous bladder rupture (SBR), in the absence of blunt or penetrating trauma, is an extremely rare urological emergency first defined in 1929 by Sisk and Wear [1].

Previous studies suggest that SBR can be a life-threatening condition, which necessitates prompt diagnosis and treatment for successful outcomes [2]. However, due to the rarity of this condition, its predisposing factors, presenting symptoms, proper diagnostic methods, and optimal treatment regimens are not well known to physicians.

In patients with SBR, underlying pathological conditions weaken the bladder wall and precipitate perforation. Risk factors known to cause

SBR include inflammation, infection, pelvic irradiation, bladder tumor, and urinary retention [2]. However, as shown in Table 1, a literature search using the terms “spontaneous bladder rupture” and “cervical cancer” in PubMed identified only 10 English articles describing a total of 15 cases of SBR in women with a history of cervical cancer [3–12]. Out of them, 14 cases were complications of pelvic radiotherapy.

Herein, we report a very rare case of SBR in a cervical cancer patient without a history of pelvic radiotherapy, which was attributable to bladder distension due to a neurogenic bladder. We also summarized current knowledge regarding cervical cancer associated SBR. This case has been reported in line with the SCARE 2020 criteria [13].

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**Table 1**  
Summary of reports of SBR developed in women with a history of cervical cancer.

Author (Year)	Reference	Age Clinical stage <sup>a</sup>	Cervical cancer treatment	Time from RT to SBR	Presenting symptoms	Initial diagnosis	Initial treatment	Delay in SBR diagnosis	Type of SBR	Possible Cause of SBR	Treatment for SBR	Recurrent SBR (Treatment)
Addar MH, et al. [3]. (1996)		40 IIB	Definitive RT	15 years	Abdominal pain Nausea/ Vomiting Hematuria	SBR	Laparotomy	0 day	Intraperitoneal	RT	Bladder repair	No recurrence
Tabaru A, et al. [4]. (1996)		74 IIIB	Definitive RT	10 years	Abdominal pain Vomiting Hematuria	SBR	Laparotomy	0 day	Intraperitoneal	RT	Bladder repair	No recurrence
Fujikawa K, et al. [5]. (1998)		57 IIIB	Definitive RT	15 years	Abdominal pain	SBR	Laparotomy	NA	Intraperitoneal	RT	Bladder repair	No recurrence
		67 IB	Definitive RT	12 years	Abdominal pain	Intestinal perforation	Laparotomy	NA	Intraperitoneal	RT	Bladder repair	No recurrence
		60 IIB	Definitive RT	10 years	Abdominal pain	Intestinal perforation	Laparotomy	NA	Intraperitoneal	RT	Bladder repair	1 year later (Bladder repair)
		48 NA	Definitive RT	10 years	Abdominal pain <b>Oliguria</b>	Peritonitis	Percutaneous drainage	NA	Intraperitoneal	RT	Bladder repair	1 week later (Bladder repair)
		68 NA	Definitive RT	13 years	Abdominal pain	Acute abdomen	NA	6 months	Intraperitoneal	RT	Bladder repair	5 months later (Ureterostomy)
		78 NA	Definitive RT	30 years	Abdominal pain	SBR	Indwelling catheter	NA	Intraperitoneal	RT	Indwelling catheter	4 months later (Bladder repair)
Nishimura T, et al. [6]. (2000)		66 IIA	Hysterectomy + adjuvant RT	3 years	Abdominal pain	Acute abdomen	NA	1 week	Intraperitoneal	RT Neurogenic bladder	Bladder repair	1 year later (Bladder repair)
Kim MK, et al. [7]. (2009)		70 NA	Hysterectomy + adjuvant RT	3 weeks	Abdominal pain Dysuria	SBR	Laparotomy	0 day	Intraperitoneal	Internal iliac artery ligations	Total cystectomy Ureterostomy	No recurrence
Snauwaert C, et al. [8]. (2012)		44 IB	RT followed by Hysterectomy	5 years	Abdominal pain	Bacterial peritonitis	Antibiotics	NA	Intraperitoneal	RT Neurogenic bladder	Bladder repair	No recurrence
Shin JY, et al. [9]. (2014)		55 NA	Hysterectomy + Adjuvant RT	13 years	Abdominal pain Distension <b>Oliguria</b>	Acute abdomen Renal failure	Hemodialysis Nephrostomy	14 days	Intraperitoneal	RT-induced urethral stricture	Bladder repair	No recurrence
Welp A, e al [10]. (2020)		27 IIIC1 (FIGO2018)	Definitive RT	0 days (Last day of RT)	Abdominal pain Nausea/ Vomiting	SBR	Indwelling catheter	0 day	Extraperitoneal	RT	Indwelling catheter (10 weeks)	No recurrence
Iwasaki H, et al. [11]. (2021)		52 NA	Hysterectomy + Adjuvant RT	7 years	Abdominal pain	Peritonitis	Antibiotics	Several weeks	Intraperitoneal	RT	Indwelling catheter (6 months)	No recurrence
Salleh, et al. [12]. (2022)		71 NA	Definitive RT	30 years	Abdominal pain Nausea	Perforated appendix	Laparotomy	0 day	Intraperitoneal	RT	Bladder repair	No recurrence
Current case		74 IB3	Hysterectomy	–	Abdominal pain Fever	Bacterial peritonitis	Antibiotics	2 weeks	Intraperitoneal	Neurogenic bladder	Indwelling catheter followed by Ureterostomy	No recurrence

NA; not available; SBR, spontaneous bladder rupture; RT, radiation therapy; FIGO, The International Federation of Gynecology and Obstetrics.

<sup>a</sup> Clinical stage of cervical cancer.

## 2. Presentation of case

A 74-year-old nulliparous Japanese woman presented with acute abdominal pain and intermittent fever. Her past medical history was unremarkable. The patient was treated with radical hysterectomy and pelvic lymphadenectomy for cervical cancer 9 days ago. The patient was unable to void postoperatively, which was attributed to a *radical hysterectomy-associated* neurogenic bladder. Subsequently, she underwent intermittent self-catheterization (6 times a day). She had no history of pelvic radiation therapy. On evaluation, she had marked guarding and rebound tenderness over the abdomen. Laboratory tests revealed a white blood cell (WBC) count of 19,330 cells/mm<sup>3</sup>, platelet count of 333,000 cells/mm<sup>3</sup>, a hemoglobin level of 9.6 g/dL, C-reactive protein (CRP) of 24.84, and serum-creatinine (Cr) of 0.92 mg/dL. Computed tomography (CT) of the abdomen and chest with contrast media suggested a small amount of intra-abdominal fluid accumulation, with no evidence of bowel obstruction or free air. Based on a diagnosis of generalized bacterial peritonitis, the patient was first treated with piperacillin and tazobactam while placing an indwelling urine catheter, which showed only mild therapeutic efficacy. Urine outflow from the vaginal stump was noted 5 days after the initiation of antibiotics. As a development of vesicovaginal fistula or SBR was suspected, we consulted a urologist for examinations. Although a cystoscopy only showed granular edematous mucosa and failed to confirm a fistula in the bladder, subsequent contrast-enhanced CT (Fig. 1A) and CT cystogram (Fig. 1B) showed a bladder wall defect with the presence of contrast medium in the abdominal cavity. With a diagnosis of SBR caused by neurogenic bladder-associated dysuria or urinary retention, the patient was initially treated conservatively with antibiotics and prolonged catheterization. However, even two weeks later, a CT cystogram revealed leakage of the contrast medium into the abdominal cavity. Subsequently, the following *treatment options* and their potential risks were discussed with the *patient and her husband*; continuation of conservative treatment, surgical bladder repair, or ileal conduit urinary diversion. Although a continuation of conservative treatment or a bladder repair was recommended by both gynecologists and urologists, the patient desired to receive a radical and curative treatment and ileal conduit urinary diversion was finally selected. With the patient's informed consent, ileal conduit surgery was performed 4 weeks after her initial presentation. The *post-operative course was uneventful*. The patient is currently healthy without further complications or cancer recurrence 4 months postoperatively.

## 3. Discussion

In this report, we present a very rare case of SBR that developed in a cervical cancer patient without a history of pelvic radiotherapy, which was attributable to bladder distension due to a neurogenic bladder.

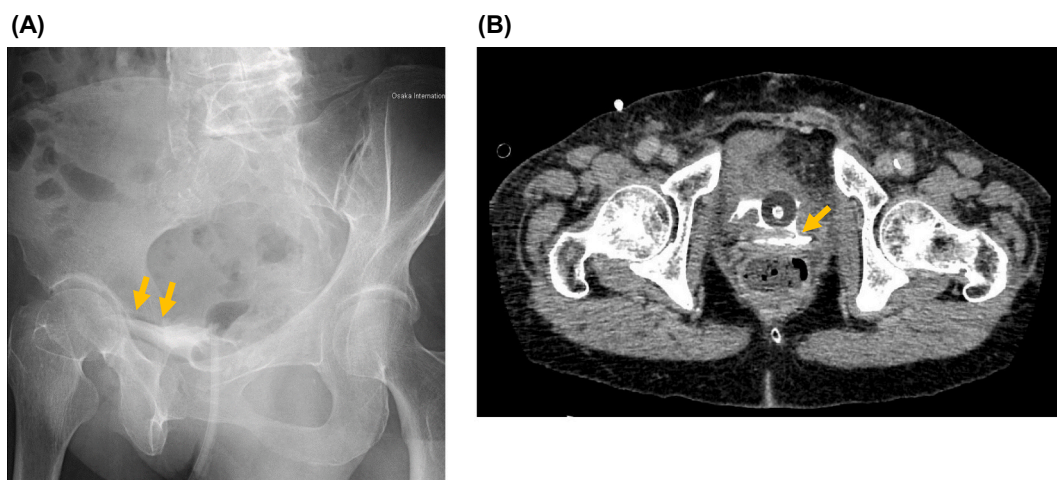
SBR is a rare but important urological emergency. SBR can cause peritonitis or sepsis, leading to poor prognosis; the mortality rate can range from 1 to 50 %, depending on the cause of SBR and the time of diagnosis [2,15].

The precise incidence of SBR remains unclear; however, a previous report estimated the incidence of SBR to be approximately 1 in every 126,000 patients [15]. However, a previous investigation from a Japanese institution suggested that approximately 2.1 % of Japanese women had bladder injury after radiotherapy for gynecologic malignancies [8]. Moreover, as shown (Table 1), 10 out of 15 reported SBR cases developed in cervical cancer patients were from Japanese institutions. Thus, it may be advisable to pay attention to SBR in Japanese women with a history of pelvic radiotherapy for gynecological malignancies.

SBR can be attributed to various predisposing factors. Common causes of SBR include inflammation, infection, bladder diverticulum, tumor infiltration, and pelvic irradiation [2]. Chronic urinary retention due to alcohol intoxication, diabetes mellitus, obstructed labor, or neuropathic bladder also predisposes a patient to bladder wall weakness and rupture [2]. As shown in Table 1, all cervical cancer patients with SBR had a history of pelvic radiotherapy. To our knowledge, our case is the first cervical cancer associated SBR without a history of pelvic radiotherapy. In this case, SBR was attributable to bladder distension due to a radical hysterectomy-induced neurogenic bladder.

The diagnosis of SBR is challenging as presentation of bladder rupture is usually non-specific, ranging from lower abdominal pain to urological symptoms of dysuria, hematuria, or urinary tract infection (UTI) [2]. Due to its low incidence and non-specific presentation, most physicians have a low index of suspicion for SBR. Therefore, the diagnosis and treatment of SBR are often delayed and missed. As shown in Table 1, among the 15 patients with cervical cancer, the most common presenting symptom was abdominal pain, and SBR was correctly diagnosed at initial presentation in only six cases.

Although previous reports have suggested that extraperitoneal rupture is more common (60–65 % of cases) and that intraperitoneal rupture occurs in 25 % of cases [14], our literature review showed that intraperitoneal rupture is predominant in women with a history of cervical cancer (14 out of 15 reported cases). Consistent with this, at the initial presentation, the majority of SBR (10 out of 15 cases) were



**Fig. 1.** Pretreatment imaging results

(A) CT cystogram: The leakage of contrast medium into the abdominal cavity was shown.

(B) Contrast-enhanced CT: A bladder wall defect and leakage of contrast medium into the pouch of Douglas were demonstrated.

misdiagnosed as generalized bacterial peritonitis, intestinal perforation, or just “acute abdomen” in women with a history of cervical cancer (Table 1).

No current treatment guidelines, based on thorough clinical evidence, exist for SBR. Extraperitoneal ruptures are usually managed conservatively with antibiotics and prolonged catheterization [15]. Surgical exploration with repair is the standard of care for intraperitoneal injuries due to the life-threatening risk associated with abdominal sepsis and peritonitis [2,15]. However, in a patient with a small perforation or multiple comorbidities unsuitable for general anesthesia, conservative treatment with antibiotics, abdominal drainage, and catheterization may be considered [2]. As shown in Table 1, among the 15 cervical cancer associated SBR cases, the treatment for SBR was surgery in 12 cases and conservative in three cases. This may be explained by the fact that intraperitoneal ruptures are predominant in women with a history of cervical cancer. In our case, conservative treatment with an indwelling catheter showed no therapeutic efficacy for 4 weeks. Although a further continuation of conservative treatment or a bladder repair was recommended by both gynecologists and urologists, the patient desired to receive a radical and curative treatment and the patient was ultimately treated surgically with an ileal conduit urinary diversion. As can be seen, even after complete resolution of the SBR, recurrent SBR was observed in 33.3 % of patients who underwent surgical treatment (4 of 12) and conservative treatment (1 of 3), indicating that there are limitations to the current surgical and conservative treatments for SBR (Table 1).

#### 4. Conclusion

We presented the first case of SBR that developed in a cervical cancer patient without a history of pelvic radiotherapy, which was attributable to bladder distension due to a radical hysterectomy-associated neurogenic bladder. It is worth noting that SBR may be present in women with surgically treated cervical cancer, even without a previous history of pelvic radiotherapy. To avoid a delay in SBR diagnosis, the condition should be considered, especially in cancer patients with a radical hysterectomy-associated neurogenic bladder exhibit an acute abdomen.

#### Informed consent statement

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Guarantor

Nariaki Matsuura, M.D., Ph.D.  
Chairman, Osaka International Cancer Institute.

#### Ethical approval status

Ethical approval is exempt at our institution.

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Not applicable.

#### CRediT authorship contribution statement

HH(Harue Hayashida), writing original draft, acquisition of data: SM (Seiji Mabuchi), conceptualization, design of the study, writing original draft: NK(Norihiko Kawamura), SM(Shinya Matsuzaki), TH(Tsuyoshi Hisa), reviewed & edited original draft: SK(Shoji Kamiura), final approval of the version to be submitted.

#### Declaration of competing interest

Not applicable.

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