

Acute Group A *Streptococcal* Pelvic Peritonitis after Radical Trachelectomy

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Abstract

Cervical cancer is the second most common malignancy in women, between the ages of 20 and 39, after breast cancer. The radical trachelectomy, developed by Daniel Dargent in 1987, is a treatment for early-stage cervical cancer in women who wish to preserve their fertility. There are few reports on pelvic peritonitis after a radical trachelectomy. Reports of Group A *Streptococcus* (GAS) infection in the field of gynecology are rare; and ascending vaginal infections have been reported in postpartum women, elderly women with low estrogen levels, and women after an uterine manipulation. In cases of severe peritonitis or endometritis, a hysterectomy might be required.

This is a case report of severe GAS pelvic peritonitis and pleurisy in a woman after a radical trachelectomy, who was not in a low estrogen state. Her condition improved temporarily with early antimicrobial treatment and drainage, but worsened again; following this, we performed a timely laparotomy with adequate cleansing and were able to preserve her uterus. Three and six months later with her follow-up appointment she was healthy and clear of infection. Women who have undergone a radical trachelectomy have a shorter cervix and potentially lower cervical defense mechanism than other women. If therapeutic intervention for severe pelvic peritonitis is delayed, then there is a possibility of losing the uterus. Obstetricians and gynecologists need to be aware of the possibility of GAS pelvic peritonitis in women who are not in a low estrogen state and should administer antimicrobial agents early. A laparotomy should be considered if there is poor improvement of symptoms.

Keywords: Peritonitis after radical trachelectomy; Group A *Streptococcus*; Vulvovaginitis; Hysterectomy

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Introduction

Cervical cancer is the fourth most frequently diagnosed cancer in women worldwide and ranks fourth in mortality [1]. Among women aged 20-39 years, cervical cancer was the second most common malignancy following breast cancer, with an incidence rate of 17.0 per 100,000 per year and a mortality rate of 9.9 per 100,000 per year [2]. The radical trachelectomy was developed by Daniel Dargent in 1987 as a treatment for early-stage cervical cancer in women wishing to preserve their fertility [3]. With regards to complications following a radical trachelectomy, there are many reports on perinatal outcomes and menstrual abnormalities, but few reports on pelvic peritonitis [4-7].

Pelvic peritonitis in women is often an ascending infection from the vagina, and the most common causative organisms are *Chlamydia trachomatis* and *Neisseria gonorrhoeae* [8]. Reports of Group A *Streptococcus* (GAS) infections in the gynecological field are rare; however, the reported mortality rate is 40% when Toxic Shock Syndrome (TSS) is caused by GAS [9,10]. GAS infections are commonly transmitted through the skin or pharynx but ascending vaginal infections have been reported in postpartum women, elderly women with low estrogen levels, and women after uterine manipulation [11-14]. If the disease progresses rapidly and causes severe peritonitis or endometritis, a hysterectomy may be required [15]. Kaiser JE, et al. [16] stated that a hysterectomy was necessary in 60% of cases of GAS peritonitis in postpartum

women with signs of capillary leakage, even if they did not satisfy TSS criteria. In this report, we describe the acute course of GAS pelvic peritonitis and pleurisy in a woman who was not in a low estrogen state. This is the first report of GAS pelvic peritonitis in a woman after a radical trachelectomy.

Case Report

A 33-year-old nullipara woman underwent a laparoscopy-assisted radical trachelectomy for stage IA2 cervical cancer (squamous cell carcinoma) at 31 years of age. The patient reported that she had sexual intercourse a day before she woke up with abdominal pain. As the pain intensified, she went to see her local physician, but the cause of her abdominal pain remained unknown. She was administered painkillers; however, the abdominal pain worsened, and she was taken to the emergency room of our hospital.

On arrival, the pain was still present, but she had clear consciousness. Her temperature was 39.2°C, pulse 130/min, and blood pressure 133/88 mmHg (Figure 1). She presented with no skin or pharyngeal symptoms; however, generalized abdominal tenderness and muscular defenses were detected. On vaginal examination, her uterus was mildly tender with a small amount of discharge present. A transvaginal ultrasound showed a small amount of ascitic fluid in the pouch of Douglas. On the ninth day after the last menstrual period, the urine pregnancy test result was negative. White Blood Cell (WBC) count, 9200/μl; C-reactive Protein (CRP) level, 0.26 mg/dl; and other laboratory results were normal. Computed Tomography (CT) showed ascites in the pelvis and inflammation in the tissue around the uterus, and no enlarged appendix or any lymphatic cysts (Figure 2). She was admitted to our hospital on account of severe abdominal pain. Five hours after admission, her pulse rate was 120/min and blood pressure 83/52 mmHg, and she went into shock. Her WBC count dropped rapidly to 2300/μl, and we suspected sepsis. She was moved to the intensive care unit and commenced on treatment for shock and an infusion of tazobactam/piperacillin administration (4.5 g every 6 h). Twenty-four hours after

admission, she developed pleural effusion, poor oxygenation, and went into acute respiratory failure (Figure 3). A thoracic drain was placed in the right lung. On the third day after admission, her vaginal secretions and blood cultures showed the presence of a GAS infection. She presented with hypotension, which satisfied the diagnostic criteria for *Streptococcal Toxic Shock Syndrome* (STSS). She also had liver damage and coagulation abnormalities; however, these did not satisfy the criteria of STSS. She was also dyspneic due to the pleural effusion but did not display the typical pulmonary symptoms of acute respiratory distress syndrome and therefore did not qualify for the diagnosis of STSS. We changed the antimicrobial agent to sulbactam/ampicillin (3 g every 6 h). Her respiratory condition improved, and the chest drain was removed on the fifth day. On the ninth day, the amount of ascites increased. Her temperature was 37.0°C, but she presented with a prolonged inflammation with a WBC count of 10800/μl and CRP level of 11.7 mg/dl; we inserted an 8 Fr drain in the pelvic floor under CT guidance. On the 18th day, she developed a rash all over her body; therefore, she was switched to cefazolin (2 g every 8 h). On the 19th day, she started menstruating, and the infection worsened again on the 20th day with right hypochondrium pain; her temperature was 38.6°C; WBC count, 12700/μl; and CRP level, 13.3 mg/dl. We changed the antimicrobial agent to tazobactam/piperacillin (4.5 g every 6 h) but discontinued it as the drug-related rash systemically spread again. We changed to meropenem (8 g every 8 h) and vancomycin, but her abdominal and chest pain as well as blood tests results worsened (WBC count: 12000/μl and CRP level: 28.4 mg/dl). Her temperature was 40°C; with worsening pleural and ascitic effusions, chest pain, and abdominal pain (Figure 4). On the 25th day, we performed a laparotomy and washed the abdominal cavity out. Three drains were placed (one each) in her vesicouterine, Douglas, and Morrison fossae. We changed the antimicrobial agent to cefazolin (2 g every 8 h), daptomycin (350 mg every 24 h), and metronidazole (1000 mg/day), and the inflammation subsided. We were able to remove the three drains after approximately 2 weeks, and she was discharged on the 47th day. Although we

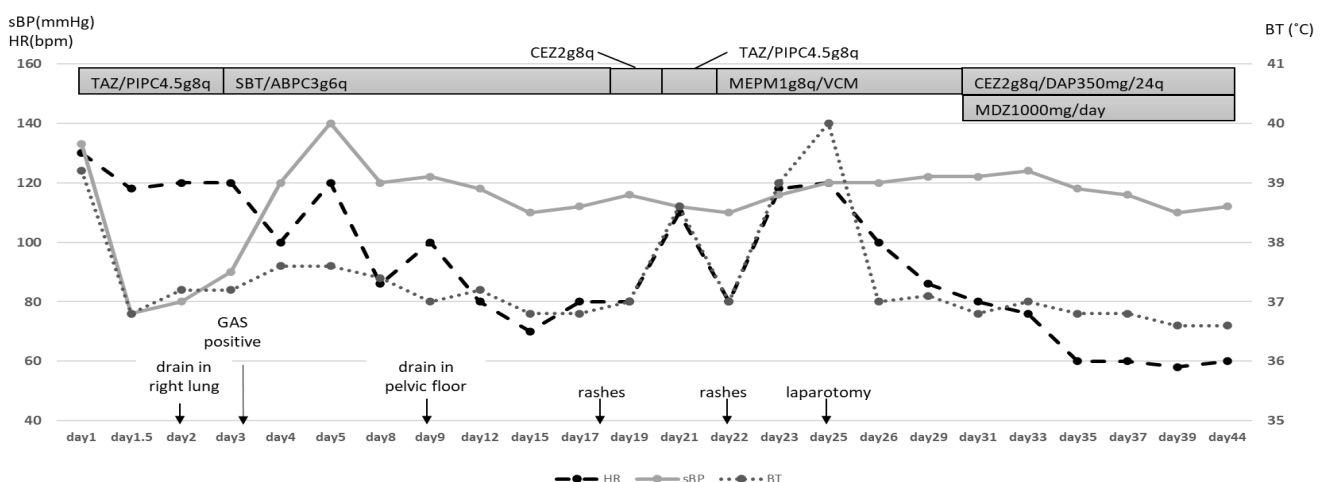


Figure 1 Clinical course.

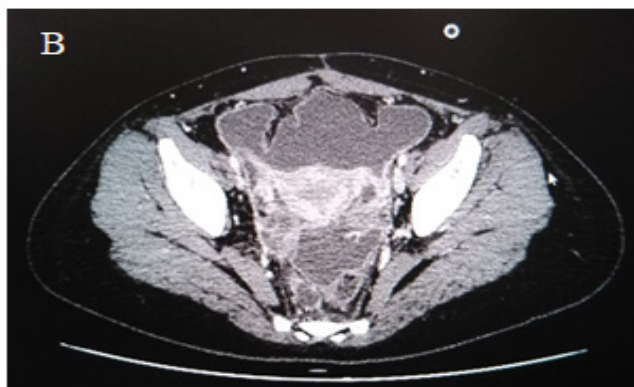
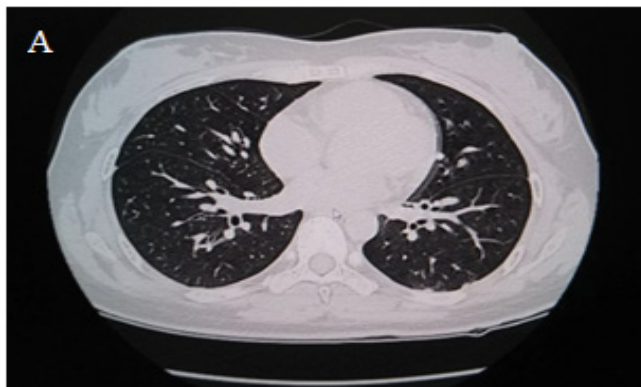


Figure 2 A) A chest computed tomography (CT) at admission and B) Abdominal CT at admission showing ascites around the uterus.

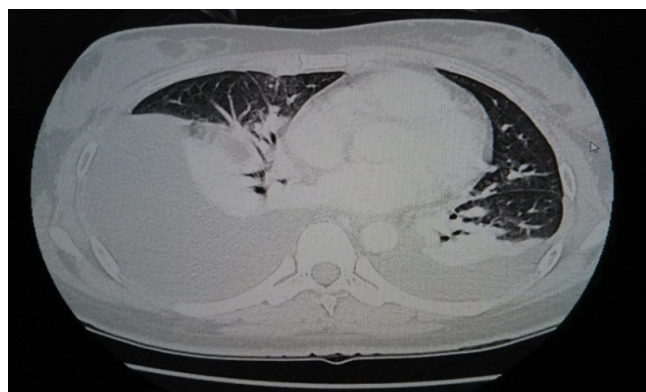


Figure 3 A chest computed tomography 24 h after admission: Showing rapidly progressive pleural effusion.

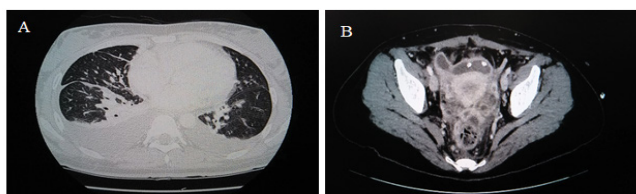


Figure 4 A) A chest Computed Tomography (CT) on the 20th day of hospitalization showing recurrent pleural effusion and worsening chest pain and B) An abdominal CT on the 20th day of hospitalization showing the pelvic floor drain *in-situ*, but with accumulating ascites.

experienced difficulty in treating her, we were able to preserve the patient's uterus after her radical trachelectomy.

Discussion

The most common routes of GAS infections are the skin and the pharynx. The occurrence of GAS vulvovaginitis has been reported more commonly in children [17], and less frequently in adult females [18]. Severe GAS infections caused by vulvovaginitis have been reported in postpartum and elderly patients with low

estrogen levels. It has been reported that about 85% of pelvic peritonitis in women is transmitted vaginally during sexual intercourse. There have been some reports of GAS infection after sexual intercourse, but they were all patients in a low estrogen state [19]. The occurrence of a GAS infection after manipulation of the uterus, for instance when an intrauterine contraceptive device was inserted, have been reported in patients who were not in a low estrogen state, but these are rare occasions [20]. The route of infection (in our patient) was thought to be sexual intercourse the day before; however, she was not in a low estrogen state and underwent no uterine manipulation. In the literature, infection during intercourse from a partner has also been reported, but at that time we were unable to confirm the symptoms of infection in the partner.

She was not in a low estrogen state; however, she had undergone a radical trachelectomy and therefore had a shorter cervix than most women. Okugawa K, et al. [7] reported 15 cases of pelvic abscesses in 12 out of 181 patients post radical trachelectomy. The time of onset ranged from 1 month to 104 months postoperatively. The causative organisms included *Escherichia coli* and 6 cases of unknown causative organisms, but no infections (in this report) caused by GAS. Although a different procedure (in many ways) from a radical trachelectomy, a cervical conization is a surgery that also shortens the cervix. The risk of preterm labor and preterm premature rupture of the membranes has been reported to be high in pregnancies after a radical trachelectomy and cervical conization [21]. In addition to a decreased mechanical support of the cervix, it has been suggested that local immunological defense mechanisms might be impaired [21]. Svare JA, et al. [21] compared women with idiopathic preterm labor, preterm premature rupture of the membranes, and normal pregnant women (without complicated pregnancies) at 26-34 weeks of gestation. The results showed that a significantly higher proportion of patients with preterm premature rupture of the membranes previously underwent cervical conization. All the types of microorganisms that were identified in the vaginas of these women were found at the same frequency in all the other groups; however, a low Lactobacillus count was found only in the group of patients who underwent cervical conization. The study

reported that previous cervical conization may compromise the antimicrobial defense mechanisms of the cervix, and therefore promote elevated microbial colony formation.

The pelvic abscess drainage and hysterectomy are treatment options for invasive GAS infections causing peritonitis. It has been suggested before that obstetricians should not hesitate to perform a hysterectomy if the uterus is inflamed. Kaiser JE, et al. [16] reported that in postpartum GAS infections, a higher pulse and respiratory rate, lower systolic blood pressure, signs, and symptoms of capillary leakage (dyspnea, cough, shoulder pain, abdominal bloating, and chest pain) at admission, were associated with adverse outcomes even if the patient did not meet the criteria for toxic shock syndrome. Signs of capillary leakage are associated with a greater need for a hysterectomy, with 20 out of 34 patients (60%) requiring a hysterectomy. Although no other reports focused on creatine kinase levels, Dehaene et al. stated that elevated serum creatine kinase may be an indicator for a hysterectomy [22]. Our patient did not satisfy the criteria for TSS; however, she presented with symptoms of capillary leakage; which improved temporarily with antimicrobial agents and drainage. These symptoms (of capillary leakage) appeared again; resulting in an immediate laparotomy, and thorough clean-out of the abdominal cavity. Although her peritonitis was severe at the time of surgery, she presented with no elevated creatine kinase, abnormal pooling, or discharge of the uterus; we therefore made the decision to preserve her uterus. With adequate flushing and drainage, her peritonitis improved, and we were able to preserve her fertility.

Conclusion

In conclusion, because our patient was post-radical trachelectomy and had a shortened cervix, she probably had a less protective mechanism against cervical infection compared to other women. Therefore, although she was not in a low estrogen state, she developed severe pelvic peritonitis and pleurisy possibly due to an ascending infection of the GAS.

Although the rate of hysterectomy is as high as 60% for patients presenting with GAS peritonitis with signs of capillary leakage—even if the criteria for TSS are not satisfied. We were still able to preserve her uterus by performing an immediate laparotomy after antimicrobial agents and abdominal drainage failed.

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