Ruptured Tuboovarian Abscess and Septic Shock with *Clostridium perfringens* in a Postmenopausal Woman

A Case Report

Amanda Wagner, M.D., Carol Russell, D.O., Jane M. Ponterio, M.D., and Joanna C. Pessolano, M.D.

BACKGROUND: *Clostridium perfringens* is the most common causative organism of gas gangrene, a necrotizing infection of soft tissue classically associated with traumatic injuries. Recently, awareness of its occurrence in spontaneous nontraumatic contexts has been increasing. The authors report an unusual case of nontraumatic/spontaneous *C perfringens* gas gangrene localized to the adnexa.

CASE: A 55-year-old woman presented with abdominal complaints and had surgery because the computed tomographic finding of air in the abdomen led to a preoperative diagnosis of perforated bowel. An infected, draining, right tuboovarian complex and infected left tube were removed. The patient had a stormy postoperative course and was ultimately diagnosed with *C perfringens* infection/sepsis; she had to be readmitted over a month after discharge for drainage of a pelvic abscess, also due to clostridial infection. The patient ultimately underwent hysterectomy with removal of remaining adnexa. The hysterectomy specimen revealed endometrial carcinoma.

CONCLUSION: *C perfringens* can cause adnexal infection in the absence of trauma, and when such cases occur, malignancy should be suspected. (J Reprod Med 2009;54:652–654)

Keywords: *Clostridium perfringens*, endometrial cancer, gas gangrene, tuboovarian abscess, adnexa uteri.

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*Clostridium perfringens* is best known for causing gas gangrene in traumatic injuries, such as battlefield wounds. Recently, spontaneous or nontraumatic *C perfringens* infection has been documented in patients with immunocompromising conditions, such as leukemia, diabetes melitis, liver cirrhosis or colon carcinoma, that increase susceptibility. The authors report a case of nontraumatic/spontaneous *C perfringens* gas gangrene localized to the adnexa.

**Case Report**

A 55-year-old, postmenopausal woman, gravida 1, para 1001, presented to our emergency department with 2 days of abdominal pain and distention. She felt generalized weakness and nausea. Past history was significant for mild asthma and breast cancer.
treated by lumpectomy. Current medications were tamoxifen and montelukast sodium. On examination she was tachycardic, afebrile and normotensive, with the abdomen markedly distended and tender to direct palpation and rebound. Abdominal plain films and a computed tomography scan of the abdomen/pelvis revealed free air within the abdomen. Exploratory laparotomy for suspected perforated bowel revealed intact bowel and bilateral adnexal masses. The right ovary was enlarged, with an open cystic cavity draining purulent material. The proximal right fallopian tube appeared normal; the distal right tube was continuous with the draining cystic cavity in the right ovary. The left fallopian tube was dilated, with chocolate-colored fluid, and the left ovary appeared to be normal. The uterus was 20 weeks’ size with multiple myomas. There were extensive adhesions involving the uterus, adnexa and pelvic sidewalls.

The patient underwent partial right salpingo-oophorectomy and left salpingectomy. Because of her critical status, time already under anesthesia and high estimated blood loss, it was decided not to perform a hysterectomy. The patient was tachycardic and hypotensive, requiring norepinephrine to maintain arterial blood pressure despite adequate volume replacement. She remained intubated and was transferred to the surgical intensive care unit (SICU). Diagnosis was ruptured tuboovarian abscess and septic shock.

Surgical pathology revealed hydrohematosalpinx (left fallopian tube) and inflammation/micro-abscess formation in the ruptured right tuboovarian cystic complex, which contained identifiable ovarian elements as well as elements consistent with infarcted fallopian tube.

SICU stay was complicated by oliguria, acute renal failure, gradually worsening anemia and thrombocytopenia (platelet counts as low as 11,000). Liver transaminases and total and direct serum bilirubin and lactate dehydrogenase were elevated. Blood urea nitrogen/serum creatinine went as high as 63/3.8. Hematology/oncology consultation was obtained regarding thrombotic thrombocytopenic purpura versus hemolytic uremic syndrome. Antithrombin antibodies were negative. The patient received a total of 6 cycles of plasmapheresis and 11 units of packed red blood cells. She was extubated on day 8 and was transferred out of the SICU on day 15. The patient developed a deep vein thrombosis in the right common femoral vein. Enoxaparin sodium injection was started at therapeutic doses, and the patient was switched to coumadin as an outpatient. Intraabdominal cultures grew C. perfringens. Blood cultures were repeatedly negative. The patient was initially treated with imipenem-cilastatin; however, this was discontinued secondary to the thrombocytopenia. She was then started on intravenous ampicillin/sulbactam and metronidazole and sent home on oral metronidazole and amoxicillin/clavulanate.

The patient was readmitted one and a half months later because of fever. Computed tomography revealed an 8-cm-diameter pelvic collection, which was drained by interventional radiology. Cultures again revealed C. perfringens. The patient was treated with intravenous meropenem and metronidazole and sent home with a peripherally inserted central catheter line for parenteral antibiotic therapy. Due to recurrent clostridial infection, the patient was scheduled for definitive therapy. Hysterectomy was performed 1 month later, with removal of the remaining adnexa. Surgical pathology of the uterus revealed endometrial carcinoma, stage IB, grade 2.

Discussion

C. perfringens is best known for causing gas gangrene in traumatic injuries. Gas gangrene is characterized by extensive local tissue destruction progressing to profound shock and death. More recently, nontraumatic incidents of C. perfringens have been documented, usually involving patients with immunocompromising conditions, such as diabetes mellitus, liver cirrhosis or malignancy (e.g., leukemia and colon carcinoma), that increase susceptibility to infection. The authors believe this is the first case report of nontraumatic/spontaneous C. perfringens infection localized to the adnexa. A PubMed search of the English-language literature turned up no similar reports.

C. perfringens is a gram-positive bacterium. The pathogenesis of gas gangrene by C. perfringens is linked to the alpha toxin that the bacterium produces, which possesses phospholipase C and sphingomyelinases, which cause the breakdown of cellular membranes. It is through this breakdown of tissue that gas is produced. Although the exact mechanism by which the bacteria that colonized the adnexa of our patient is not known, it is important to keep in mind certain characteristics of C. perfringens. C. perfringens thrives in areas with poor vascularity, anaerobic conditions and acidic conditions. The bacterium’s ability to increase vascular perme-
ability allows its toxins to reach, and affect, large areas.

The finding of endometrial carcinoma in the hysterectomy specimen is of interest given the association of nontraumatic \textit{C perfringens} infection with malignancies.\textsuperscript{3,4} Possible mechanisms that could predispose a patient with endometrial malignancy to develop clostridial infection include: (1) a relatively oxygen-poor, acidic environment due to malignant tissue outgrowing its blood supply; (2) a relative deficit in the number of cellular immune responders on the basis of the postulated outgrowth of blood supply; and (3) altered local immune response due to some defect in, for example, defensin or other antimicrobial peptide production or function. Given that \textit{C perfringens} is commonly found in the gastrointestinal tract,\textsuperscript{6} any alteration in endometrial immune response and oxygenation could allow, after inadvertent autoinoculation of the vagina with clostridia, a greater chance of the bacteria ascending and infecting the adnexa.

This patient was at an increased risk for endometrial cancer because of her history of tamoxifen use. She had not seen a gynecologist during this time. She denied any history of postmenopausal bleeding and had not had endometrial surveillance while on tamoxifen.

\textit{C perfringens} infections can cause myriad symptoms that make the exact diagnosis difficult. It is important to rapidly identify involved tissue for excision. Since early infection is typically asymptomatic or nonspecific, \textit{C perfringens} is often brought to clinical attention at an advanced stage. Prompt, aggressive management of \textit{C perfringens} is essential in improving patient survival. However, the decision to perform extensive procedures is frequently complicated by an unstable clinical condition. Mortality from \textit{C perfringens} gas gangrene remains high, ranging from 67\% to 100\%.\textsuperscript{7}

\textit{C perfringens} can cause adnexal infection in the absence of trauma. When such cases occur, malignancy should be suspected.

\textbf{References}