Research Letter

An atypical and fatal case of pyometra accompanied by the superficial spread of squamous cell carcinoma of the endometrium and the fallopian tubes

Anne Chao a, Alex Ming C. Wang b, Tzu-Hao Wang c, Tzu-I. Wu d, An-Shine Chao c,*

a Department of Anesthesiology, National Taiwan University Hospital, National Taiwan University, Taipei, Taiwan
b Department of Radiology, Chang Gung Memorial Hospital, Chang Gung University, Taoyuan, Taiwan
c Department of Obstetrics and Gynecology, Chang Gung Memorial Hospital, Chang Gung University, Taoyuan, Taiwan
d Department of Obstetrics and Gynecology, Wan Fang Hospital, Taipei, Taiwan

Accepted 30 May 2013

Pyometra, an accumulation of large amounts of purulent material within the uterine cavity, is an uncommon condition that carries a significant risk of morbidity and mortality in elderly women with underlying medical conditions [1–5]. We herein report an extremely rare case of a postmenopausal woman with unruptured pyometra who presented with unusual clinical, imaging, and microbiological findings. The persistence or progressive accumulation of purulent discharge (1200 mL) led to a massive uterine enlargement, which was evident on ultrasound and computed tomography (CT) images. The patient ultimately died from septic shock while she was undergoing uterine drainage. Her blood cultures demonstrated a heavy growth of Staphylococcus epidermidis. Notably, the histopathological examination of the uterus showed the presence of squamous cell carcinoma in situ arranged in a diffuse pattern that had spread superficially into the entire endometrial cavity and the fallopian tubes. Moreover, the patient had multiple foci of invasive squamous cell carcinoma scattered along the myometrium. The upward superficial spread of squamous cell carcinoma extending over the entire surface of the endometrium is a very uncommon clinical presentation (0.1%) [6,7].

A 60-year-old, para 3, woman had noted increasing abdominal girth for approximately 2 months. Upon admission she presented with abdominal distension and a palpable abdominal mass. The patient was in good general conditions and did not suffer a net weight loss of significant proportions. She had no fever and her white blood cell count was normal (5300/μL). The tumor marker carcinoembryonic antigen was significantly increased (20.97 ng/mL), whereas cancer antigen-125 levels were in the normal range (25.6 U/mL). Physical examination revealed a nontender pelvic mass without gross lesions in the vagina or exocervix. The patient had negative Pap smears. Transabdominal ultrasound imaging revealed the presence of a hypoechogenic fluid-filled abdominal mass (22 cm × 16 cm × 10 cm in size) with echogenic debris, compatible with a diagnosis of pyometra (Fig. 1A). Transvaginal ultrasound showed a marked dilatation of the cervical canal with an upward extension into the uterine cavity. A CT imaging revealed the presence of a cervical structure (Fig. 1B) as well as a marked enlargement of the uterine cavity (19 cm × 15 cm × 13 cm in size), which was filled with relatively high-density fluid. Moreover, the patient had multiple small-sized bilateral lung lesions, which were suggestive of pulmonary metastases. Intravenous administration of cefazolin (1 g) was started prior to attempting uterine drainage under anesthesia. The evacuation of a large amount (1200 mL) of purulent material from the uterus was followed by the sudden onset of shock. Upon performing emergency laparotomy, the abdominal cavity was found to be clean and there was no evidence of uterine perforation. We then attempted an emergency subtotal hysterectomy and bilateral salpingooophorectomy. Moreover, the patient began receiving an antibiotic regimen consisting of vancomycin, cefazidime, and metronidazole. Unfortunately, she died 2 days later because of septic shock and multiple organ failure caused by persistent bacteremia. Two of her three cultured blood samples showed a heavy growth of S. epidermidis. A gross pathological examination revealed a smooth and enlarged endometrial cavity covered with a layer of whitish mucosa. A histopathological analysis showed that the endometrium was entirely covered by...
a diffuse squamous cell carcinoma in situ, which extended both to the cutting edges of the cervical tissue and to the lumen of the fallopian tubes bilaterally. We also identified invasive foci (3 mm in depth) of moderately differentiated squamous cell carcinoma that penetrated into the myometrium, approximately 20% of myometrial thickness. We also tested formalin-fixed, paraffin-embedded tissues for human papillomavirus (HPV) structural antigens and DNA types (King Car HPV blot). All of the endometrial and fallopian tube specimens tested positive for HPV-16.

Pyometra is a serious medical condition characterized by the accumulation of purulent material in the uterine cavity resulting from interference with its natural drainage. Recognition and aggressive management of this condition are essential because it still carries significant risk of morbidity and mortality [1]. Several factors that increase the risk of pyometra have been identified, including viral or bacterial cervicitis, radiation exposure, the presence of foreign bodies in the uterine cavity (e.g., intrauterine devices), exposure to chemical agents, estrogen treatment, vitamin A deficiency, and intracavitary tumors [1,5]. It is noteworthy that pyometra can be the first sign of an underlying neoplasm; malignancies are the underlying cause of this condition in 3.5—45% of all cases [2].

Most postmenopausal women with pyometra present with one or more of the following symptoms: vaginal discharge, vaginal bleeding, abdominal pain, and fever [1—3]. The presence of a palpable abdominal mass containing more than 500 mL of purulent material is an uncommon finding in postmenopausal patients with pyometra [1—3]. Moreover, this condition is generally caused by Bacteroides fragilis, Streptococcus spp., or Escherichia coli [1—3]. Our patient had at least two uncommon and noticeable features, namely: (1) the accumulation of a large amount of purulent material (1200 mL) in the uterine cavity in the absence of typical clinical symptoms; and (2) the presence of S. epidermidis isolates from blood cultures. S. epidermidis is a type of bacteria commonly found on the skin and in the vaginal mucosa but is rarely a cause of clinical infections. Our patient most likely had a hematogenous spread of S. epidermidis from an infective focus in the lower genital tract. Antibiotics for patients with pyometra should cover both aerobic and anaerobic organisms, especially in the preoperative period. Clinicians should be aware of the potential risks of infectious complications and sepsis during uterine drainage. Immediate laparotomy, peritoneal lavage, and even hysterectomy are indicated in the presence of suspected perforation of pyometra, as in our case. Unfortunately, these procedures carry a high risk of morbidity and death, especially in elderly patients with severe underlying medical conditions [4,5]. Ultrasound is an essential diagnostic tool for both pyometra and cervical malignancies. In our patient, ultrasound suggested the presence of an intrauterine disease by showing a cervical stenosis in the absence of bulky cervical masses. The intrauterine superficial spread of a cancerous endometrial lesion may be evident on gross inspection as whitish patches. This pattern has been referred to as “cake icing” or “zuckerguss” carcinoma, a condition that occurs when the superficial squamous cell carcinoma sweeps over or replaces the normal healthy endometrium [6,7]. In our patient, the presence of a cervical stenosis and the subsequent pyometra may have had a facilitating role in the promotion of the superficial spread of a cervical malignancy as well as the endometrial extension of an endometrial cancer. Our case showed a histological continuity of the malignant spread from the endocervix, through the endometrium, to the fallopian tubes. By contrast, we found no downward superficial spread of the malignancy to the vagina. The HPV infections are known to play a key role in the pathogenesis and clinical outcomes of cervical and endometrial squamous cell carcinomas [8]. All of the specimens tested in our case were positive for HPV-16 infection, suggesting the HPV persistence and clinical evolution of squamous cell carcinoma in this patient. Primary endometrial squamous cell carcinoma with extensive local spread and pyometra is generally characterized by concomitant tubal involvement. By contrast, isolated intrauterine metastases from primary squamous cell carcinoma of the
fallopian tube are rare [9,10]. The endometrial involvement by endometrial cell carcinoma does not alter the International Federation of Gynecology and Obstetrics clinical staging system. However, the prognosis of squamous cell carcinoma spreading to the surface of the endometrium has been occasionally reported to be poorer than that of typical endometrial cell carcinoma of the endometrium [6,7,9,10].

In conclusion, we presented a rare and fatal case of pyometra with an atypical presentation and high morbidity-associated pathology that should be taken into consideration.

References