Inversion of the Uterus Combined with Endometrial Carcinosarcoma

Huan-Huan Zhao, Jian-Xin Cheng, Li Li

Department of Obstetrics and Gynecology, The Forth Hospital of Hebei Medical University, Shijiazhuang, Hebei 050011, China

To the Editor: A uterine carcinosarcoma (malignant mixed Müllerian tumor [MMMT]) is a rare, aggressive, malignant tumor which demonstrates both malignant epithelial (carcinoma) and mesenchymal (sarcoma) components.[1] MMMTs account for 2–5% of all uterine malignancies. Metastatic progression is uncommon and occurs through hematologic, lymphatic, and intraperitoneal spread. Risk factors for MMMTs include radiation, excessive estrogen exposure, obesity, and nulliparity. The 2-year survival rate for patients with MMMTs is 53% for Stage I disease and decreases to 8.5% for Stages II and III.[2]

Uterine inversion involves a trapped uterine fundus, which in turn causes the endometrium to protrude through the cervix to the vagina. Uterine inversion can be divided into the following three types: (1) incomplete uterine inversion, in which the uterine fundus descends inferiorly but does not pass through the external cervical os; (2) complete uterine inversion, in which the fundus and corpus extend through the external os; and (3) total uterine inversion, in which the vagina is also inverted.[3] In the process of diagnosis and treatment, uterine inversion patients usually have combined symptoms of severe pain, bleeding, infections and shock labor performance, the precondition of which include relief pain, controlled hemorrage, infection and shock. The differential diagnosis included the submucous myoma of uterus and uterine prolapse, vaginal wall cyst, or vaginal wall prolapsed. Lupovitch A, *et al*.[4] reported a rare case of uterus inversion happened in endometrial sarcoma of the uterus. Nonpuerperal uterine inversion with endometrial carcinosarcoma is an unusual condition but can occur in the postmenopausal age group. Clinical diagnosis of the disease is difficult and can sometimes be fatal. This study introduced a case of uterus inversion combined with endometrial carcinosarcoma.

A 74-year-old Chinese woman complained of irregular vaginal bleeding for 6 months and a vaginal mass for 30 days. Due to urination and gravity, the vaginal mass descended to the vulva. The mass was 10 cm × 8 cm × 7 cm in size and accompanied by substantial vaginal bleeding. A physician in another hospital performed uterine artery embolization and biopsied the mass, which was shown to be a uterine carcinosarcoma. The vaginal hemorrhage decreased significantly postoperatively. The medical history was unremarkable and she had no prior surgeries. There was no family history of uterine myomas or malignancies.

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The patient came to our hospital for further evaluation. The vaginal examination revealed a large polypoid mass that was thought to be a prolapsed uterus [Figure 1]. Pelvic ultrasonography revealed no uterine echo in the pelvic cavity. A magnetic resonance imaging (MRI) of the pelvis showed that the uterus and carcinosarcoma had prolapsed through the vagina. The tumor markers were normal with the exception of CA-125 (607.20 U/ml). The primary diagnosis was endometrial carcinosarcoma combined with inversion of the uterus. For treatment, a laparotomy was planned. Because of the hypercoagulable state (D-dimer >17.2 mg/L), the patient was given a low-molecular-weight heparin sodium injection twice daily until the D-dimer level approached normal. The inverted uterus and vulva were cleaned daily. To avoid intraoperative bleeding, artery embolization was repeated on the day before surgery. Exploration during laparotomy showed that the uterine

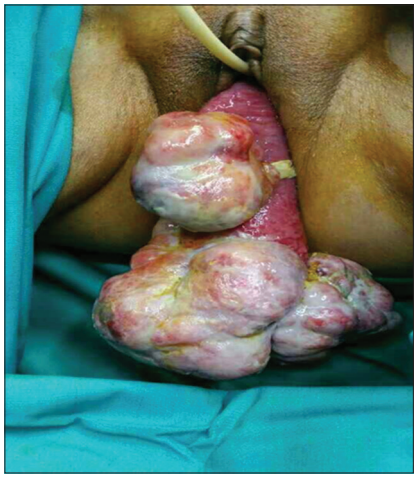

Figure 1: The inverted uterus with tumor prolapsed to the vulva. No anatomic features of a uterus are seen externally.

Address for correspondence: Dr. Li Li,
Department of Obstetrics and Gynecology, Forth Hospital of Hebei Medical University, Shijiazhuang, Hebei 050011, China
E-Mail: lily_lucky1@163.com

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corpus was absent. The uterine adnexa were visualized with multiple nodules on the pelvic floor and greater omentum. After the endometrial carcinosarcoma tissue was resected, the uterus was restored to the anatomic position, and a hysterectomy with bilateral salpingo-oophorectomy was performed. The greater omentum was resected as well. The postoperative pathology showed that carcinosarcoma cells had invaded into the myometrium, cervical canal, pelvic peritoneum nodules, and sigmoid colon mesangial nodules. Immunohistochemistry revealed the following: ER (1+), PR (1+), P53 (0), Ki-67 (20–80%+), CK (+), Vim (+), CD10 (+/−), Act (+/−), Des (+), S-100 (+), and CK79 (+).

The patient declined radiotherapy and chemotherapy; thus, no postoperative treatment was rendered. Carcinosarcoma spread throughout the body after 5 months, and the patient had difficulty urinating and defecating. Two months later, the patient died.

Inversion of the uterus is a rare condition which is an obstetric emergency and a diagnostic challenge for gynecologists. Nonpuerperal uterine inversion is often associated with leiomyomas, sometimes associated with uterine sarcomas, but very rarely associated with endometrial carcinosarcoma. A typical presentation of carcinosarcoma combined with uterus inversion includes pyometra with vaginal bleeding, bloody or watery discharge, abdominal pain, or a polypoid mass in a postmenopausal woman. In such cases, it is easy to misdiagnose a uterine carcinosarcoma as a cervical tumor.

Uterine inversion is suspected when a tumor is present in the vagina, but the uterine fundus is not palpable by pelvic examination. It is noteworthy that an MRI or computed tomography (CT) scan is a useful tool in the diagnosis of nonpuerperal inversion of the uterus. In the present case, the inverted uterus and endometrial carcinosarcoma were observed in the vulva. The vagina was inverted as well, which is rare and consistent with a total inverted uterus. Ultrasonography demonstrated no uterine corpus in the pelvis. An MRI examination showed a total inverted uterus and a malignant mass involving the endometrium. The etiologic factors leading to an inverted uterus and carcinosarcoma include (a) sudden extrusion of a tumor from the uterus, (b) thin uterine wall, (c) dilatation of the uterine cervix, and (d) tumor size. The primary treatment or uterine carcinosarcoma with uterine inversion is surgical, including total hysterectomy, bilateral salpingo-oophorectomy, bilateral pelvic lymphadenectomy, and omentectomy.[5,6] The final histologic evaluation confirmed the diagnosis of Stage IIIb carcinosarcoma. The behavior of carcinosarcomas is characterized mainly based on the carcinomatous component, thus carcinosarcomas metastasize to the lymph nodes. The benefit of the role of lymphadenectomy was apparent. In our case, the pelvic and para-aortic lymph nodes were not palpable, and because of comorbidities and the physical condition of the patient, a lymphadenectomy was not performed.

Radiotherapy is suggested in the literature as another option for the treatment of uterine malignancies. In general, it is known that the 5-year survival of patients with uterine carcinosarcomas receiving radiotherapy versus no irradiation was 41.5% and 33.2%, respectively.[7] A retrospective study assessed the efficacy and toxicity of a novel combination chemotherapy using carboplatin, ifosfamide, and mesna compared with other regimens for patients in adjuvant and palliative settings. Indeed, combined adjuvant radiotherapy and chemotherapy may increase the 5-year survival rate.[8]

Declaration of patient consent

The authors certify that they have obtained all the appropriate patient consent forms. The guardians have given their consent for patients images and other clinical information to be reported in the journal. The guardians understand that patient's names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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