

# A case of uterine incarceration caused by carcinosarcoma accompanied with pyometra

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## Summary

Uterine incarceration is rare clinical condition which principally occurs after the first trimester during pregnancy. Main clinical symptoms of uterine incarceration are abdominal pain and urinary symptoms such as urinary retention. Until now, almost all cases of uterine incarceration have been associated with the gravid uterus. To the present authors' knowledge, only a few reports of this condition associated with uterine leiomyoma have been reported in the English literature. They present the first case of uterine incarceration caused by malignant gynecologic neoplasm (carcinosarcoma) accompanied with pyometra. Inferring from the patient's symptoms and vaginal examination findings, they clinically diagnosed her as uterine incarceration, thereafter magnetic resonance imaging revealed characteristic findings consistent with uterine incarceration. The clinical course of the present case and clinical considerations regarding uterine incarceration are discussed.

**Key words:** Carcinosarcoma; Pyometra; Retroverted uterus; Urinary retention; Uterine incarceration.

## Introduction

Incarceration of the gravid uterus is known as a rare but serious complication during pregnancy [1]. This condition occurs when the retroverted uterus remains impacted within the hollow of the pelvic cavity and cannot ascend out of the pelvis as the pregnancy advances [2]. The incidence of uterine incarceration is one in 3,000 to 10,000 pregnancies in the second trimester of pregnancy [3]. Clinical presentation includes severe abdominal pain, pelvic pain, and urinary symptoms such as retention, dribbling, and incontinence [4]. However, asymptomatic cases have often been unrecognized throughout pregnancy until just before delivery and can lead to serious obstetric emergencies such as dystocia, uterine rupture, fetal demise, and maternal unintended organ injury during cesarean section [2, 5].

To the present authors' knowledge, almost all previously reports regarding uterine incarceration have been associated with the gravid uterus in the English literature. There are only three reports of non-gravid uterine incarceration associated with leiomyoma published in 1989, 2007, and 2012 [6-8].

The authors present the first case of uterine incarceration caused by malignant gynecologic neoplasm accompanied with pyometra. The clinical course of our case and clinical considerations concerning this rare condition are discussed.

## Case Report

A healthy 56-year-old woman, gravida 2 para 2, visited this hos-

pital because of postmenopausal genital bleeding for two months, and abdominal pain with peritoneal signs and occasional urinary

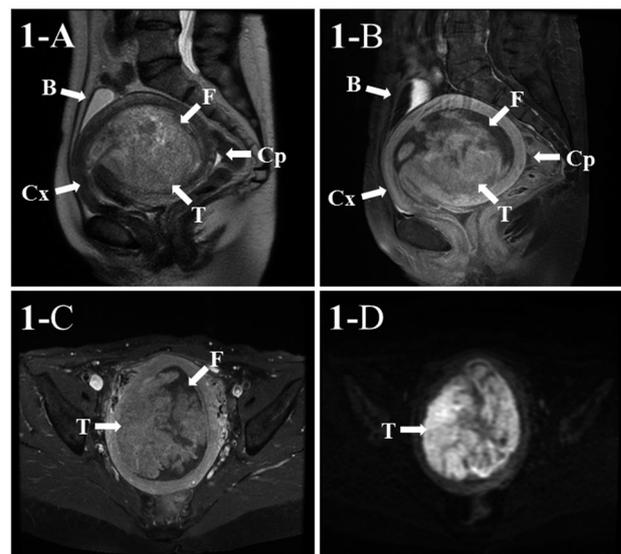


Figure 1. — Findings of preoperative magnetic resonance imaging. (1-A: sagittal T2-weighted image, 1-B: sagittal contrast-enhanced T1-weighted image, 1-C: transverse contrast-enhanced T1-weighted image, and 1-D: transverse diffusion-weighted image). Magnetic resonance imaging clearly reveals severely retroverted uterine corpus (Cp) fully-filled with a papillary growing tumor (T) accompanied with a heterogeneous fluid collection (F) suggestive of pyometra, an anteriorly displaced and elongated uterine cervix (Cx), and a markedly upward extended bladder (B). The strong enhancement and the higher ADC values of the tumor indicate the possibility of uterine malignancy with tumor necrosis.

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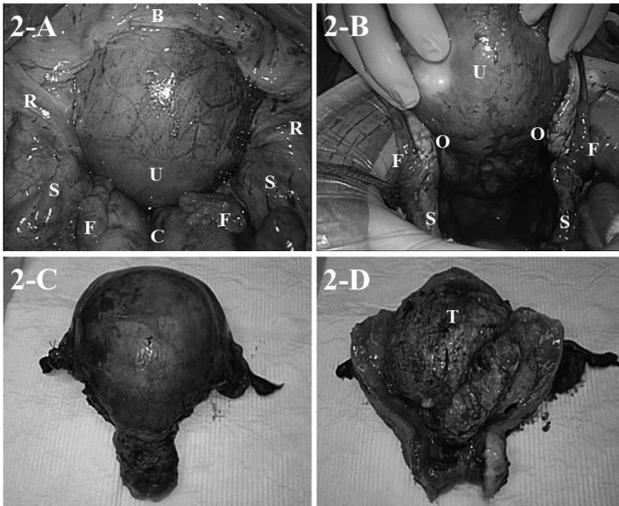


Figure 2. — Intraoperative findings and the macroscopic findings of isolated specimen. 2-A: Severely retroverted uterus (U) is tightly compacted into the pelvic cavity. Bilateral round ligaments (R) and suspensory ligaments of ovary (S) are drawn into the pelvic floor area. Bilateral ovaries (O) and the fundus of uterus are completely invisible. (B: bladder, C: sigmoid colon, and F: fallopian tube). 2-B: After uplifted the enlarged uterus out from the pelvic cavity, no adhesion is observed around the internal genital organs. 2-C: The shape of the uterus is just like a gravid uterus. 2-D: The uterine cavity is filled with a polypoid necrotic tumor (T) and pus.

retention for a week. Vaginal examination showed a small amount of bloody pus-like discharge and an extreme displacement of the uterine cervix lifted up behind the pubic bone. Transvaginal ultrasonography showed a 10-cm enlarged retroverted uterine corpus with an indeterminate intrauterine pathological lesion and ventrally displaced uterine cervix. Inferring from her symptoms and vaginal examination findings, the authors clinically diagnosed her with uterine incarceration. Magnetic resonance imaging (MRI) clearly revealed severely retroverted uterine corpus fully-filled with a papillary growing tumor accompanied with a significant heterogeneous fluid collection suggestive of pyometra, an anteriorly displaced and elongated uterine cervix, and a markedly upward extended bladder (Figure 1). She had a fever (body temperature: 39.0°C) and blood testing showed her acute infectious condition and anemia; white blood cell count: 9,800 (4,000-8,000 / $\mu$ L), hemoglobin: 8.6 (11.5-15.5 mg/dL), and C-reactive protein: 9.95 (< 0.30 mg/dL). Therefore, she was diagnosed with uterine incarceration caused by an indeterminate gynecologic tumor accompanied with pyometra and pelvic inflammatory disease. She was immediately treated by surgery. At laparotomy, her retroverted uterus was tightly compacted into the pelvic cavity. (Figure 2). Bilateral round ligaments and suspensory ligaments of the ovary were drawn into the pelvic floor area. Therefore, bilateral ovaries and the fundus of uterus were completely invisible. However, the authors could smoothly relieve the uterine incarceration because no adhesion was present around the internal genital organs and the consistency of the uterine corpus was deformable and soft. After they uplifted the enlarged uterus out from the pelvic cavity, hysterectomy, and bilateral salpingo-oophorectomy were uneventfully performed. The pathological diagnosis confirmed homologous carcinosarcoma accompanied with pyometra. (Figure 3). Her med-

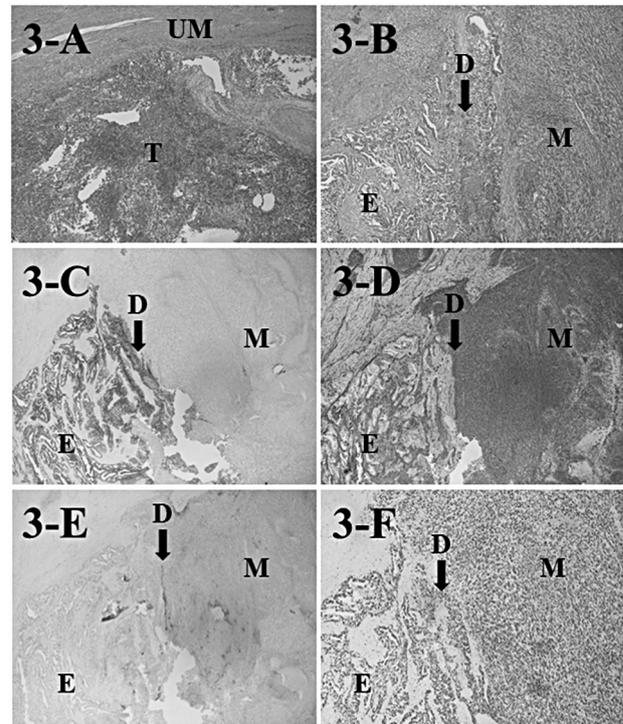


Figure 3. — Histopathological findings of the tumor. 3-A: Malignant tumor (T) is deeply invasive into the uterine myometrium (UM). 3-B: The tumor is composed of a mixture of malignant epithelial and mesenchymal elements, namely grade 3 endometrioid adenocarcinoma and endometrial stromal sarcoma. There is a sharp demarcation (D) between the epithelial element (E) and mesenchymal elements (M). Immunohistochemical findings demonstrate positive immunostaining for cytokeratin (3-C) and vimentin (3-D) and negative for CD-10 (3-E) with epithelial element (E), positive immunostaining for vimentin (3-D), and CD10 (3-E) and negative for cytokeratin (3-C) with mesenchymal element (M), and positive immunostaining rates for MIB-1 (3-F) being over 40%. These findings were compatible with homologous carcinosarcoma.

ical treatment was followed by additional surgery (pelvic and para-aortic lymphadenectomy) and adjuvant chemotherapy with paclitaxel and carboplatin.

## Discussion

In 11% of pregnant women the gravid uterus was retroverted in the first trimester [9]. In most cases, the gravid uterus moves towards an upward position before 14-16 weeks' gestation and keeps this position throughout the rest of pregnancy [5]. However, when the uterus remains retroverted as the pregnancy progresses, the growing gravid uterus remains trapped between the sacral promontory and pubic symphysis, and then uterine incarceration may develop [2]. The same mechanism can explain the pathogenesis of the present case of uterine incarceration. Uterine carcinosarcoma aggressively grows as a large, soft, polypoid mass filling, and distending

the uterine cavity [10]. Tumor necrosis and hemorrhage are prominent features of uterine carcinosarcoma and they are likely to form a hotbed of intrauterine bacterial infection [10]. According to the polypoid tumor growth and the formation of pyometra, the uterine corpus is rapidly enlarged just like the shape of the gravid uterus. Moreover, if the uterus is comparatively retroverted, the intrauterine accumulated pus fluid is hard to drain out through the cervical canal, which accelerates the further enlargement of pyometra and increases the risk of developing uterine incarceration.

The type of complication associated with uterine incarceration varies with the causes of this condition or the stage of pregnancy [5]. In the first trimester of pregnancy, the most commonly reported complication is acute urinary retention [4, 11]. The symptoms of abdominal pain and urinary symptoms are a consequence of the compression of the bladder trigone by the anteriorly displaced uterine cervix [1, 8]. In the present case, the size of the uterus is just the same size of late first trimester of pregnancy; therefore clinical symptoms are typical of uterine incarceration with a similar size gravid uterus. In the second and third trimesters of pregnancy, intrauterine fetal death, intrauterine growth retardation, premature labor and delivery, renal failure, and sepsis have previously been reported with uterine incarceration [1, 5]. At delivery, which inevitably requires cesarean section, an unintentional iatrogenic injury of maternal bladder, cervix, and/or vagina can occur because of the anterosuperior positioning of these organs with uterine incarceration. [1, 2, 9]. Complications of uterine incarceration are serious and potentially devastating for both mother and fetus [5].

Early diagnosis and detailed investigation are crucial for the management of uterine incarceration [9]. Diagnosis of uterine incarceration is clinical and confirmed by imaging [1]. First, knowledge of this rare condition is important [5]. This case report will contribute to increase awareness regarding uterine incarceration among all clinicians of both obstetrics and gynecology. Second, all clinicians should be aware of the signs and symptoms of uterine incarceration. Women presenting with abdominal pain and urinary symptoms, such as urinary retention should have a vaginal examination performed to exclude uterine incarceration [11]. A vaginal examination would reveal the ventralized vaginal axis and the elongated anteriorly displaced and poorly visualized uterine cervix, which should trigger an awareness of uterine incarceration and a further evaluation [4]. Third, the characteristic findings of MRI play a key role in diagnosing uterine incarceration [4, 5, 9]. MRI is superior to ultrasonography, not only for accurate diagnosis but to clearly elucidate the patients' distorted pelvic anatomy [1]. Surgery for uterine incarceration (especially during cesarean section) is technically challenging even in cases of known uterine incarceration [2]. Therefore, preoperative surgical simulation based on the information obtained from the MRI image is critical for avoiding an inadvertent organ injury during surgery [5, 8].

## Conclusion

The present authors experienced a rare case of uterine incarceration caused by carcinosarcoma accompanied with pyometra. The possibility of uterine incarceration should be considered in any patients with symptoms of abdominal pain and urinary retention. When the diagnosis is suspected on the basis of clinical presentation or a vaginal examination, characteristic MRI findings can enable definitive diagnosis of uterine incarceration. Appropriate clinical assessment and management should be done expeditiously to avoid increased adverse patient (both maternal and fetal during pregnancy) outcomes.

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