



Abstract

Chronic non-puerperal uterine inversion is an uncommon gynecological condition that is frequently misdiagnosed as a prolapsed submucosal leiomyoma, often resulting in delayed diagnosis and management. We report the case of a 41-year-old woman presenting with a six-month history of persistent metrorrhagia and severe anemia, who was subsequently diagnosed with a grade 3 uterine inversion caused by a large, prolapsed endometrial mass. Management involved initial exploratory laparoscopy, followed by conversion to laparotomy, culminating in a total hysterectomy with bilateral salpingo-oophorectomy. Histopathological analysis confirmed high-grade endometrial stromal sarcoma. This case underscores the diagnostic complexity and therapeutic challenges associated with chronic uterine inversion, particularly when linked to underlying malignancy.

Keywords: Uterine Inversion; Non-Puerperal; Endometrial Stromal Sarcoma; Management; Diagnosis; MRI.

Introduction

Uterine inversion, characterized by the turning inside out of the uterus, is a rare but serious condition more commonly seen in obstetrics than gynecology [1]. This condition, also referred to as "uterine invagination," poses significant morbidity due to the risk of hypovolemic shock [2]. It is classified into four degrees based on the extent of fundal displacement: first-degree, with the fundus forming a concavity within the endometrial cavity; second-degree, where the fundus descends through the external cervical os; third-degree, involving prolapse of the fundus into the vaginal canal or exteriorization at the vulva; and fourth-degree, including inversion of both the uterus and vaginal walls [3]. This rare presentation is often misdiagnosed, with a prolapsed submucosal leiomyoma being the primary differential diagnosis. We present a case of chronic non-puerperal uterine inversion associated with third-degree genital prolapse, emphasizing the diagnostic and therapeutic challenges involved.

Case Report

A 41-year-old patient presented with six months of persistent metrorrhagia, accompanied by fatigue and symptoms of severe anemia. She was single, nulligravid, and had no significant medical history, although her sister had been diagnosed with endocervical adenosarcoma. No genetic testing had been performed, and no psychosocial barriers to healthcare access were identified. The patient had previously been hospitalized twice for severe anemia, with a hemoglobin level as low as 2.4 g/dL, necessitating the transfusion of four units of packed red blood cells. Initial management with tranexamic acid and progestin therapy failed to resolve her symptoms.

Upon admission, her vital signs included a blood pressure of 90/60 mmHg and a heart rate of 100 bpm. Physical examination revealed marked conjunctival pallor, and gynecological examination showed a distended hymen due to a vaginal mass with

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mild bleeding. A rectal examination confirmed a bulging vaginal mass. Initial pelvic ultrasound and MRI identified a 7×5 cm uterine corporo-isthmic mass with signs of hemorrhagic necrobiosis. An initial biopsy suggested angiomyxoma, but further biopsy findings showed nuclear atypia of the cytogenic stroma, which were inconclusive for malignancy. A re-evaluation of the pelvic ultrasound revealed the absence of the uterus in its normal anatomical position and the presence of a hypervascular pelvic mass distending the vaginal canal, along with an additional vascularized round pelvic mass. A subsequent MRI confirmed a grade 3 complete uterine inversion, that was caused by a large, suspicious endometrial mass, characterized by marked diffusion restriction and a type 3 enhancement pattern. The mass had prolapsed into the vaginal cavity. An additional mass in the left cervico-isthmic region, likely involving the left ovary, was noted (Figure 1).

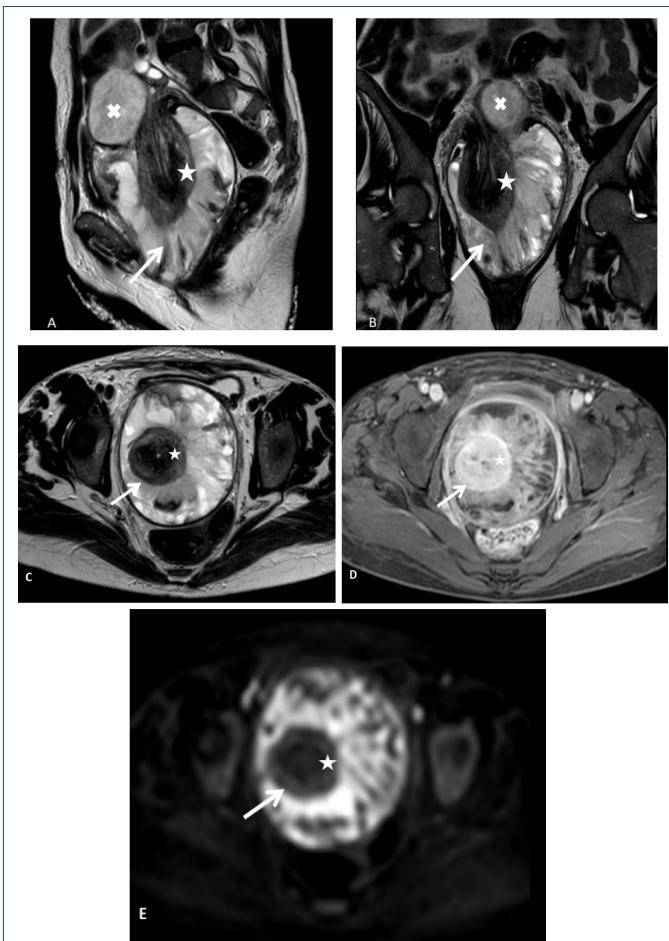


Figure 1: Pelvic MRI shows a U-shaped uterus (astérix) with prolapse of the uterine fundus into a distended vaginal cavity, corresponding to grade 3 uterine inversion. This inversion is best visualized on sagittal T2-weighted (A) and coronal T2-weighted (B). The inversion is caused by a large endometrial mass (arrow), which shows high signal intensity with heterogeneous internal architecture on the axial T2-weighted (C) images and restricted diffusion on the diffusion-weighted axial image (E, $b=1000$). Post-contrast T1-weighted axial images with fat suppression (E) reveal a suspicious enhancement pattern (type 3). A characteristic “ring appearance” resulting from the arrangement of serosa, myometrium and endometrium. The cross mark reveals an additional supracervical mass, probably involving the left ovary.

Under general anesthesia, an exploratory laparoscopy confirmed the uterine inversion. While the right adnexa were visualized, the left adnexa were entrapped within the inversion. Due to the complexity of the case, a laparotomy was performed via a midline incision. Repeated attempts to correct the uterine

inversion vaginally were unsuccessful and resulted in significant vaginal bleeding, necessitating bilateral ligation of the hypogastric arteries. A total hysterectomy with bilateral salpingo-ooophorectomy was subsequently performed using a combined abdominal and vaginal approach. Final histopathological analysis confirmed a high-grade endometrial stromal sarcoma. Despite initial stabilization, the patient developed metastatic disease and passed away one year postoperatively.

Discussion

Chronic non-puerperal uterine inversion is a rare and complex condition that poses significant diagnostic and management challenges in contemporary clinical practice. Although uterine inversion is commonly associated with the postpartum period, non-puerperal cases are exceedingly uncommon [2]. This condition primarily affects postmenopausal women or those over 45 years of age [1]. The clinical presentation varies based on the degree of uterine inversion: first-degree involves a concave indentation in the vaginal fornix, second-degree sees the uterine fundus passing through the external cervical os, third-degree presents with the fundus prolapsed into the vaginal canal or occasionally exteriorized, and fourth degree includes involvement of the vaginal walls [3]. Two additional classifications further describe uterine inversion: one based on the extent of inversion (partial or complete) and the other on its progression (acute or chronic). While acute non-puerperal uterine inversion is typically characterized by severe pain and heavy menorrhagia, chronic inversion is marked by pelvic discomfort, minimal vaginal bleeding, and significant anemia [4].

Several predisposing factors contribute to the development of non-puerperal uterine inversion, including a thin uterine wall, a large or poorly anchored tumor, cervical dilation due to uterine cavity distension, and tumor dehiscence. Submucosal fibroids represent the most common etiology, accounting for 70-85% of cases, while malignant tumors such as leiomyosarcomas, embryonal rhabdomyosarcomas, and endometrial stromal sarcomas are responsible for 15-30% [5]. Diagnosis relies on a combination of clinical examination and imaging. A thorough vaginal examination should assess the position of the cervical os and the depth of the vaginal cul-de-sacs to distinguish uterine inversion from prolapse. In cases of complete uterine inversion, the cervical os may be located at the vaginal introitus, complicating identification [6]. Rectal examination often reveals a cup-shaped depression, a hallmark of uterine inversion. Imaging plays a crucial role in the diagnosis of uterine inversion. Ultrasound, CT scans and MRI can all provide valuable diagnostic information. Uterine inversion can be difficult to diagnose on the basis of ultrasound alone. It may reveal a vaginal or cervical mass on longitudinal views, with the appearance of an inverted uterine fundus and on a target sign on axial views [7]. CT is not generally used as a primary diagnostic tool for uterine inversion. It can be used in emergencies or for staging an underlying malignancy.

MRI is the key imaging modality for the diagnosis of uterine inversion, as it not only confirms the diagnosis and determines the degree of inversion, but also identifies the underlying cause. On sagittal images, MRI typically reveals a U-shaped uterine configuration with loss of the normal convexity of the uterine fundus. Axial images often show the characteristic “ring sign”, resulting from the variable signal intensities of the inverted uterine layers (serosa, myometrium, endometrium and vagina). In addition, MRI is highly effective in detecting associated underlying pathologies, such as submucosal fibroids or malignant

endometrial tumours [5, 6].

Management of non-puerperal uterine inversion depends on the underlying cause, patient health, and the severity of symptoms. Conservative approaches, such as manual uterine reduction, are typically considered for hemodynamically stable patients with favorable anatomical conditions [8]. The success of this method depends on factors such as the patient's anatomy, the cause of the inversion, and timely intervention. To prevent recurrence, a pessary may be employed following successful reduction [9].

When conservative methods fail or are not suitable, surgical intervention becomes necessary. Hysterectomy is often the preferred treatment, particularly when fertility preservation is not desired. Among the various surgical techniques, the Hultain procedure is considered the most effective according to the literature [8, 10]. The choice between vaginal and abdominal approaches depends on factors such as the surgeon's expertise, patient anatomy, and specific uterine characteristics (9). Additional considerations, such as coexisting pelvic pathologies and fertility preservation, play a crucial role. Vaginal procedures, however, can be technically demanding, especially in the absence of uterine prolapse.

The potential for perioperative complications, including bleeding or injury to adjacent organs such as the ureters, bladder, and intestines, highlights the need for surgical expertise. Postoperative care is essential, involving close monitoring for complications like infection or recurrence to ensure optimal outcomes [11].

Conclusion

Chronic non-puerperal uterine inversion is an exceptionally rare and complex condition that necessitates heightened clinical awareness for timely diagnosis and effective management. Achieving successful outcomes relies on a thorough understanding of the underlying etiology, careful diagnostic evaluation, and tailored treatment strategies. While conservative management may be suitable in specific cases, surgical intervention, particularly the Hultain procedure, is often necessary when fertility preservation is not a priority. The technical complexities and associated risks emphasize the need for surgical expertise and

meticulous postoperative care to minimize complications and ensure optimal outcomes. Increased recognition and reporting of such cases are critical to advancing diagnostic accuracy, refining management protocols, and improving patient care in the future.

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