

NON-PUERPERAL TOTAL UTERINE INVERSION DUE TO A LEIOMYOMA: A CASE
REPORT

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ABSTRACT

Introduction: Uterine inversion is an extremely rare gynecological complication, that characterized by the folding of the fundus into the uterine cavity reaching the cervix or beyond the cervix. It is most commonly associated with benign tumors such as submucosal leiomyomas. It's an extremely rare gynecological condition which is an obstetric emergency and a diagnostic challenge in gynecology. There should be high index of suspicion in the setting of a mass protruding from the vagina, non-palpable uterus, and pelvic organs not visualized on ultrasound.

Case Presentation: We report the case of a 50-year-old woman, gravida four para four, admitted to the emergency's department for an irregular vaginal bleeding, pelvic pain and externalized mass through the vulva evolving for 3 days. A total interannexial hysterectomy was performed via dual approach with vaginal route supplemented by laparotomy. Histopathological examination confirmed uterine inversion originating from a fundal myoma with no malignancy. The postoperative course was uneventful and she was discharged without complication. **Discussion:** Uterine inversion, where the uterus turns inside out by descending through the cervix, predominantly affects older women outside of childbirth, often due to uterine malignancies in younger patients. Diagnosis poses challenges, as it mimics other conditions and varies in severity from incomplete to total inversion. Imaging, such as ultrasound and MRI, aids in diagnosis. Treatment typically involves hysterectomy, although conservative options are considered for certain cases. Surgical techniques vary based on the extent of inversion and patient-specific factors, aiming to restore normal anatomy and function while addressing underlying pathology.

Conclusion: Non-puerperal uterine inversion represents a severe complication. It is most commonly associated with benign tumors such as leiomyomas, however the possibility of an underlying malignancy should not be neglected. Total hysterectomy using a dual approach via laparoscopy and vaginal access is a reliable and safe surgical technique.

KEYWORDS: Uterine inversion, Non-puerperal uterine inversion, leiomyoma.

INTRODUCTION

Uterine inversion is an extremely rare gynecological complication, that characterized by the folding of the fundus into the uterine cavity reaching the cervix or beyond the cervix. There are two types of uterine inversions including: puerperal (obstetric) and non-puerperal (gynecologic).^[1] Puerperal inversion is a serious and rare complication, which generally occurs in the third stage of the delivery with a reported incidence of roughly 1 in 15000 deliveries.^[1] Non-puerperal inversion occurs chiefly when the uterus acts to expel a fundal submucous leiomyoma but it's generally associated with a pathological mass such leiomyomas, endometrial polyps, uterine sarcomas, endometrial carcinomas, and mixed mesodermal tumours. It can be acute or chronic.^[2] Non-puerperal uterine inversion accounts for approximately 17% of all uterine inversions and less than 200 cases of non-puerperal uterine inversions have been reported in a literature review.^[2]

We present a case of a spontaneous chronic uterine inversion in which the implicated tumour was a sessile fibroid.

Our work has been reported in line with the SCARE Guidelines 2023 criteria.^[3]

CASE REPORT

A 50-year-old woman, gravida 4, parity 4, with no notable medical or surgical history of note, was admitted to our hospital's emergency ward with complaints of severe pelvic pain and abundant metrorrhagia evolving for 5 days. On admission, she was hemodynamically unstable with a blood pressure of 80/40 mmHg and pulse rate of 130 beats per minute. On examination she was pale, and there was a large, firm hemorrhagic mass filling the vagina and protruding to 10 cm beyond the introitus and the normal uterine cervix contour could not be visualized. Blood tests revealed a severe anemia with

a hemoglobin level of 4.9 g/dl. Ultrasonography showed an empty pelvis without the uterus, and non-puerperal uterine inversion was highly suspected.

Given the abundant bleeding and the unstable vital signs, four units of packed cell were transfused. Surgical management was chosen with a dual approach: vaginal route and laparotomy. We started by vaginal myomectomy to reduce the volume of the mass protruding from the vagina, and to allow the reposition of the uterus by applying gentle upward pressure vaginally. A total adnexal hysterectomy was performed via laparotomy. The abdominal approach provided not only good surgical exposure but also excluded any digestive or urinary content trapped in the inversion before hysterectomy. The initial workup on the first postoperative day showed: hemoglobin level of 8.9 g/dl.

The postoperative course was uneventful, and the patient was discharged on the third postoperative day. Histopathological examination confirmed uterine inversion originating from a fundal myoma with no malignancy.

DISCUSSION

Uterine inversion refers to a descent of the uterine fundus to or through the cervix, so that the uterus is turned inside out. In general, non-puerperal uterine inversion presents after 45 years.^[1] The cases that presented in the reproductive age were mostly associated with a malignant tumor of the uterus.^[4] The major challenge in non puerperal uterine inversion is in making the diagnosis as it has to be distinguished from cervical and vaginal masses, and a prolapse of the uterus depending on the stage.

Uterine inversion can be classified into incomplete, complete, or total and can be categorized into 4 degrees, depending on the extent of the inversion of the fundus into four stages as described by Salomon et al. based: Stage 1: The inverted uterus remains within the uterine cavity; Stage 2: Complete inversion of the fundus through the cervix; Stage 3: The inverted fundus protrudes from the vulva; and Stage 4: Inversion of the uterus and vaginal wall through the vulva.^[5]

Benign uterine masses, such as fundal leiomyomas or endometrial polyps, have been described in the literature as causes of uterine inversion^[6]; uterine malignant tumors such sarcoma, mixed Müllerian tumor, endometrial and cervical carcinoma, rhabdomyosarcoma and immature teratoma can also cause inversion, or it can be idiopathic.^[7] A 2018 systematic review of 170 case reports identified benign leiomyomas as the primary cause of chronic uterine inversion at 57.2% followed by leiomyosarcomas at 13.5% and 9.9% of all cases showed no evidence of abnormalities.^[8] A recent literature review found malignancy in approximately 33% of the reported cases.^[9]

The major factors that contribute to its occurrence are: rapid tumor growth, a fundal location of the tumor, a small tumor pedicle, a thin uterine wall, weakness of the uterus or cervix, uterine contractions, or distension of the uterine cavity.^[8] Factors that increase intra-abdominal pressure, such as coughing, sneezing, straining, and ascites caused by chronic liver disease, also appear to be implicated.^[10]

Two modes of occurrence are described in the literature: Acute non-puerperal uterine inversion and chronic non-puerperal uterine.^[8] The acute form is characterized by a higher intensity of the pain and a heavy menorrhagia; while the chronic form is responsible of pelvic pain resembling heaviness, vaginal discharge, metrorrhagia, anemia, urinary symptoms complicated in some cases by the obstruction of the urethra, and the presence of a vaginal mass are also reported^[8], this form can also be asymptomatic; diagnosis is then radiological.^[8]

Clinical diagnosis of uterine inversion is difficult unless the fundal depression can be palpated in bimanual examination, and the cervix cannot be visualised after the vaginal mass was excised.^[6] Rectal examination is a complementary diagnostic examination of great value in order to confirm the absence of the uterus in the pelvis and to rule out elemental diagnostic doubts in more severe uterine inversions.^[11] Unfortunately, because of the rare nature of the disorder, uterine inversion frequently goes undetected until surgery unless a high index of suspicion is maintained.

Imaging is essential for diagnosis, Ultrasound examination is the first line imaging investigation, a U-turn sign, showing a central course of the main uterine vessels instead of their normal anatomical peripheral location laterally alongside the corpus of the uterus, may represent a novel and pathognomonic sign of uterine inversion.^[12] The 3D power Doppler has been used more recently in the diagnosis of uterine inversion as it can clearly show the changes in the uterine artery course in relation to the uterus.^[13] Magnetic Resonance Imaging remains the best imaging modality for the diagnosis of uterine inversion, and for characterizing and delimiting the underlying lesion while studying surrounding structures, the features will include: "U" shaped uterine cavity, thickened and inverted uterine fundus on sagittal section and "bull's eye" configuration on an axial Image.^[13]

Management often involves a hysterectomy and this can be done either through the vaginal or abdominal route depending on the size of the mass, the stage at presentation and the histological diagnosis.; however, in cases of incomplete inversion or nulliparous patients requiring preservation of fertility, conservative treatment may be pursued after histopathology confirms the benign nature of the mass.^[14]

For stage 1 or 2, an attempt should be made to manually reduce the inversion to its correct anatomical position through the vagina using the Kustner technique, which involves entering the pouch of Douglas vaginally and splitting the posterior aspect of the uterus and cervix to reinvert the uterus. Alternatively, the Spinelli technique can be used, which involves making an incision on the anterior aspect of the cervix and then repositioning the uterus. Additionally, from above during a laparotomy, the Huntington technique can be employed, which involves placing a clamp on each round ligament or myometrium. On the inverted fundus, repeat the procedure until the inversion is corrected. The Haultain procedure, which involves making an approximately 1.5-inch incision on the posterior surface to transect the constriction ring, appears to be the most successful method for achieving repositioning according to a literature review. If necessary, a hysterectomy may follow.^[15-16] Stages 3 and 4 can be managed with a vaginal hysterectomy as was done in this case.^[15]

Robotic and laparoscopic surgeries and abdominal cerclage have been recently used to correct chronic uterine inversion.^[5]

In our case, surgical management was chosen with a dual approach: vaginal myomectomy followed by routine laparotomy hysterectomy. The abdominal approach, in addition to providing good surgical exposure, allowed for the exclusion of any digestive or urinary content entrapped within the inversion prior to the hysterectomy. Anatomical distortions must always be considered and anticipated during surgery to avoid complications.

CONCLUSION

Non-puerperal uterine inversion is an exceptional and life-threatening disease with a diagnosis that is challenging. It is usually associated with uterine pathology. This case highlights that nonpuerperal uterine inversion should be included in the differential diagnosis when a patient presents with protruding vaginal mass and bleeding. Careful examination, imaging studies such as pelvic ultrasound and MRI coupled with a high index of suspicion will yield correct diagnosis and treatment that would help to decrease patient morbidity and mortality.

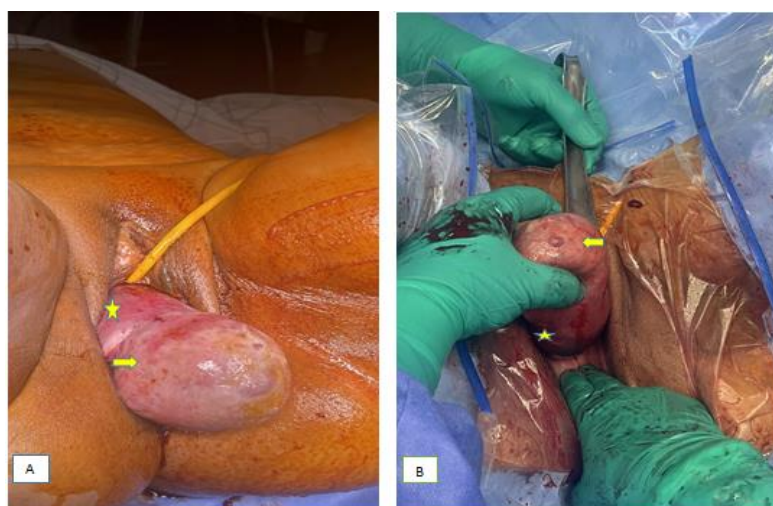


Figure 1: Complete uterine inversion due to prolapsed myomas seen through the vaginal route: A: Anterior view, B: Posterior view Inverted uterine fundus with exposed endometrium (star), Large myoma abutting from the uterine fundus (arrow).

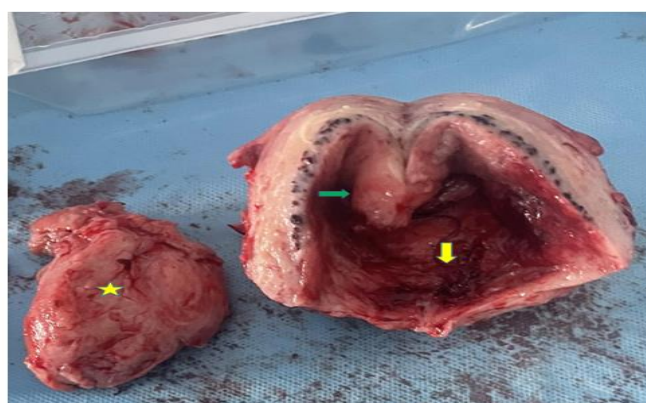


Figure 2: After hysterectomy, opening of the cavity and intra-cavitary pedunculated fibroid at the origin of the inversion: Uterine cavity (yellow arrow), Myoma pedicle (green arrow), leiomyoma (star).

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