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Case Report

# Nonpuerperal vaginal hemorrhage secondary to complete uterine inversion: A case report☆,☆☆,★,★★

*Mana Modares, BMSc, MD(C)*[a], *Josée Sarrazin, MD, FRCPC*[b,*]

[a] *Faculty of Medicine, University of Toronto, Toronto, Ontario, Canada*
[b] *Department of Medical Imaging, Sunnybrook Health Sciences Centre, University of Toronto, 2075 Bayview Avenue, Toronto M4N 3M5 Ontario, Canada*

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ABSTRACT

Approximately 95% of uterine inversion cases are associated with pregnancy in the early postpartum period. This case describes a rare presentation of uterine inversion in the nonpuerperal period secondary to a submucosal leiomyoma. A 48-year-old G2P2 perimenopausal female was admitted for 6 weeks of abnormal uterine bleeding and a 17 × 10 cm mass prolapsing into the cervical canal and upper vagina, with a large vascular pedicle inserting into the central superior aspect of the lesion from the fundal region. A computed tomography (CT) scan confirmed the diagnosis of a complete uterine inversion secondary to a large fundal leiomyoma with a submucosal component. Laparoscopic total hysterectomy was performed with no complications, and pathology confirmed the diagnosis of a benign leiomyoma. Though rare, uterine inversion can be caused by a leiomyoma in the nonpuerperal period and should be considered in patients with abnormal uterine bleeding and pelvic masses. Ultrasonography and CT scan were sufficient in providing an accurate diagnosis for which surgical management was indicated in this case.



## Introduction

Uterine inversion is a life-threatening medical emergency where the uterine fundus prolapses into the endometrial cavity, endocervical canal, and even into the vagina [1,2]. Patients commonly present with severe lower abdominal pain, a mass protruding through the cervix, and uterine bleeding. Prompt recognition of symptoms is critical as hemorrhage may be severe, leading to shock, and ultimately, death

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* Corresponding author.
E-mail address: mana.modares@mail.utoronto.ca (M. Modares).

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[1,2]. Upwards of 95% of uterine inversion cases are associated with vaginal deliveries in the early postpartum period [3,4]. The other 5% of novel, less-documented cases are nonpuerperal in nature. We present a rare presentation of a nonpuerperal complete uterine inversion in a patient with a large submucosal leiomyoma.

## Case presentation

A 48-year-old Gravida 2, Para 2 peri-menopausal female presented with a 6-week history of vaginal bleeding with an acute increase in volume over a 24-hour period. The patient was not in immediate distress on initial assessment, and review of systems was unremarkable; although, she noted feeling more fatigued compared to baseline. The patient had regular menstrual cycles of 28 days up until the initiation of abnormal uterine bleeding. At that time, she had an abdominal ultrasound, which demonstrated a 7 × 7 cm submucosal leiomyoma protruding into the endometrial cavity. Conservative management was pursued. She had had 2 previous uncomplicated spontaneous vaginal deliveries and no recent pregnancies. The patient was otherwise healthy, and she was not on any medications.

On physical exam, the patient's abdomen was soft and non-tender with her uterus palpable below the umbilicus. A sterile speculum examination demonstrated a mass protruding through the cervix, into the upper vagina. There were few necrotic areas showing slow active bleeding. A bimanual examination demonstrated a fully dilated cervix with a smooth mass protruding through the external cervical os into the upper vagina. The uterus remained mobile. A transabdominal and transvaginal ultrasound was conducted, which demonstrated a 17 × 10 cm mass prolapsing into the cervical canal and upper vagina, with a large vascular pedicle inserting into the central superior aspect of the lesion from the fundal region (Fig. 1a-c). This was followed by a computed tomography (CT) scan for surgical mapping, which confirmed the concern for complete uterine inversion secondary to a large fundal leiomyoma with a submucosal component (Fig. 2a-c). The patient was vitally and hemodynamically stable with a hemoglobin of 130g/L. Due to ongoing blood loss, a decision was made for hospital admission and hysterectomy.

During the first night of admission, the patient's hemoglobin fell from 130g/L to 107g/L over one hour due to severe vaginal hemorrhage. She was started on 1 g cyclokapron IV q8h. The patient stated that she was a Jehovah's Witness, and therefore refused red blood cell transfusion or alternative blood products, but she did consent to use of cell saver for intraoperative blood conservation. After hemodynamic stabilization, the patient was booked for immediate hysterectomy. The postoperative diagnosis following laparotomy with total abdominal hysterectomy was a necrotic pedunculated submucosal fundal leiomyoma and complete uterine inversion. The pathology report confirmed a partially necrotic leiomyoma with thrombosis and no significant cervical abnormalities. No malignancy was identified. Informed consent was obtained for publication of this patient's unique presentation.

## Discussion

Patients with uterine inversion are at risk of rapid clinical deterioration due to catastrophic vaginal hemorrhage. Therefore, prompt recognition and management become critical to establish hemodynamic stabilization in a timely manner. A case report by Al Qahtani in 2018 found that abdominal ultrasound may be inconclusive in acute settings, especially if there is complete uterine inversion or if the pedicle of the leiomyoma cannot be located precisely [5]. Therefore, magnetic resonance imaging (MRI) was used in this case for characterization of the uterine lesion [5]. This report contrasts our case and a case by Krissi et al. in 2011 [6], in which abdominal ultrasound was able to accurately demonstrate features of uterine inversion (Fig. 1a-c). Zohav et al. in 2020 introduced the diagnosis of uterine inversion through 3-dimensional power Doppler of the uterine arteries; however, further research is required to determine the efficacy of using the "U-turn" for diagnosing this life-threatening condition [2].

Upon a comprehensive review of the literature, uterine inversion in a nonpuerperal setting due to leiomyomas are quite uncommon and have variable presentations. Lima et al. in 2018 described a chronic nonpuerperal uterine inversion case in a patient who was completely asymptomatic [7]. Conversely, Della Corte et al. in 2019 described a case of uterine inversion secondary to a fibroid in a woman with similar symptoms to our case, including vaginal bleeding and fatigue [8]. A case by Singh and Ghimire in 2020 described a patient with 15 months of chronic vaginal bleeding, who finally presented acutely with hypotension and systemic shock, and was ultimately diagnosed with uterine inversion [9]. This case contrasts our patient as Singh and Ghimire describe a long course of abnormal uterine bleeding, whereas our patient had a subacute course (6-weeks) of abnormal uterine bleeding and subsequently presented acutely with complete uterine inversion [9]. A case by Dadgar and Pourhosseini in 2018 was most similar to our patient, where they describe a 51-year-old multiparous female with abnormal uterine bleeding [10]. This patient was found to have a large intramural fibroid of 5.5 × 6.2 cm and was ultimately diagnosed with uterine inversion [10]. This spectrum of patient symptomatology renders nonpuerperal uterine inversion an important diagnosis to consider in the differential for abnormal uterine bleeding and abdominal pain, particularly in the context of known uterine fibroids.

Prompt surgical treatment for this pathology is critical to reduce further blood loss and decrease morbidity and mortality. A case by Banaschak et al. in 2014 reported a case of fatal postpartum air embolization following emergency laparotomy, manual compression, and packing of the uterus due to uterine inversion [11]. This case demonstrates the severity of this presentation and importance of immediate definitive management through hysterectomy. The 2020 report by Singh and Ghimire suggests an immediate myomectomy and Haultain's procedure as an alternative to hysterectomy to preserve the uterus and child-bearing capability of the patient [9]. Haultain's procedure involves bisecting the myometrial constriction ring through posterior uterine incisions to prevent inversion [9]. Al Qahtani in 2018

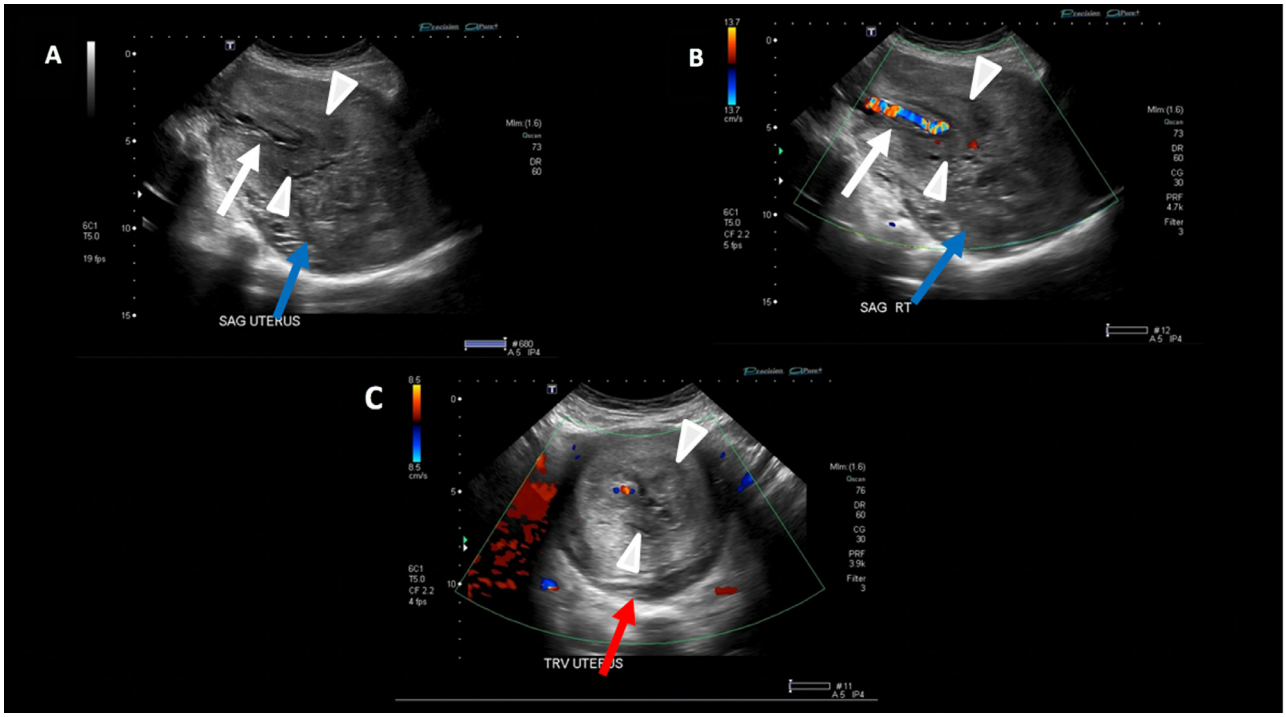


**Fig. 1 – Transvaginal ultrasound images (A) Sagittal image demonstrating a large heterogeneous central uterine mass protruding through the cervix into the upper vagina, similar to an "intussusception" appearance. (B) Sagittal image demonstrating a prominent central vascular pedicle extending from the fundal region. (C) Transverse image at the level of the uterine mid-body confirming a similar large mass with vascular core. *White arrows designate fibroid central stalk and white arrowheads demonstrate surrounding endometrium/endocervical mucosa (the latter only discernable on ultrasound) *Red arrow demonstrates vaginal wall. *Blue arrow designates fibroid leading to the uterine inversion.**

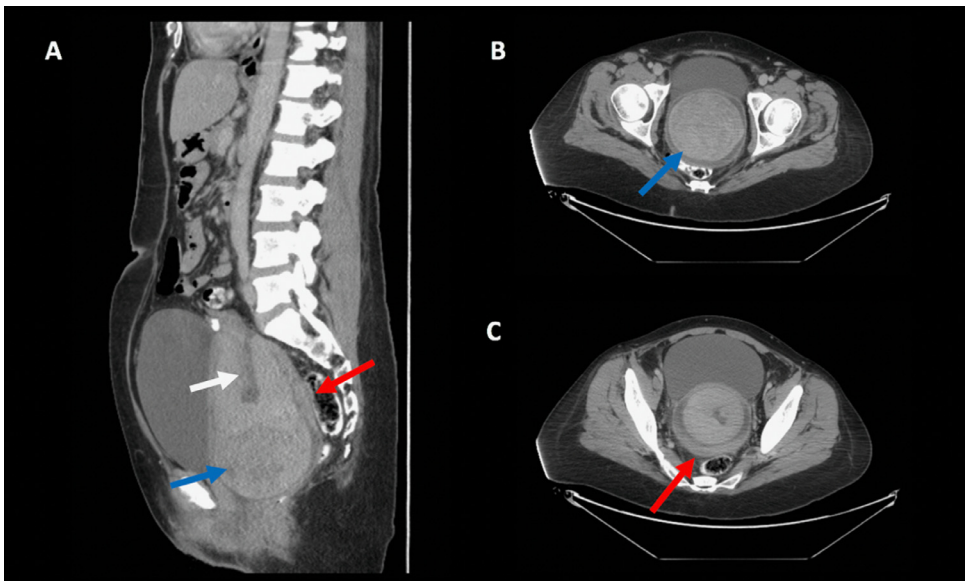


**Fig. 2 – Contrast enhanced CT images (A) Sagittal image demonstrating enlarged inverted uterus due to a large fundal mass (heterogeneously hypodense) which is the lead point for inversion, favored to represent a submucosal fibroid. (B and C) Axial images at the level of the inverted fundal mass and uterine body, respectively, demonstrating similar findings. *White arrows designate fibroid central stalk. *Red arrow demonstrates vaginal wall. Blue arrow designates inverted uterus**

described his case's successful surgical management with a vaginal approach with LigaSure [5]. Similar to the management of our patient, De et al. in 2019 treated their patient's submucosal myoma with a total laparoscopic hysterectomy with successful surgical outcomes [3]. Although our patient consented to a total hysterectomy, the gynecologists elected for a laparoscopic approach to minimize blood loss given the patient was a Jehovah's Witness. This method demonstrated good efficacy and no postoperative complications.

Alternative etiologies for uterine inversion, such as neoplastic causes, must be considered in the nonpuerperal setting. Lupovitch et al. in 2005 presented a case of a 26-year-old with uterine inversion secondary to uterine sarcoma [12]. Occhionero et al. described a similar case of a uterine inversion secondary to Müllerian uterine adenosarcoma in a 62-year-old patient [13]. In contrast, Gomez-Lobo et al. in 2008 published a case of an adolescent female with uterine inversion secondary to immature teratoma [14]. Lastly, Moulding and Hawnaur presented a case of nonpuerperal uterine inversion due to endometrial carcinoma in a 52-year-old [15]. These cases demonstrate the importance of accurate diagnosis through imaging and clinical examination to efficiently rule out malignant pathology.

In summary, we present a rare case of nonpuerperal complete uterine inversion secondary to a large submucosal leiomyoma in a multiparous patient presenting with a 6-week history of subacute abnormal uterine bleeding followed by an acute presentation of increased blood loss upon admission. Her Jehovah's Witness status complicated clinical decision making, however utilization of cell saver and a laparoscopic approach to a total hysterectomy were effective in improving outcomes and minimizing blood loss. Abdominal ultrasound was a useful imaging modality for describing the anomaly, and findings were confirmed with CT. In our case, an MRI was not required. The differential for abnormal uterine bleeding and sharp abdominal pain should be expanded to include uterine inversion, even in patients who are not postpartum, and particularly if there is a known history of uterine fibroids.

## Patient consent statement

Patient provided informed written consent for anonymized use of their data in this case report.

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