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Acute Presentation of Nonpuerperal Uterine Inversion Following a Diagnosis of Uterine Carcinosarcoma: A Case Report

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ABSTRACT

Nonpuerperal uterine inversion (NPUI) due to endometrial carcinosarcoma can present in an acute manner with life-threatening complications. The rare presentation of uterine inversion outside of the puerperium caused by a malignant etiology, which was further complicated by the urgent need for intervention, makes this case crucial in recognizing the importance of expeditious management of NPUI. A 73-year-old nulliparous woman was referred to our gynecological oncology service following a diagnostic hysteroscopy and biopsy, which had identified a 10 cm polyp that was reported to be a high-grade serous carcinosarcomatous polyp. Only 20 days after the initial diagnosis was made, the patient experienced an intensification of her symptoms with an increased volume of per vaginal bleeding, oliguria, and vomiting. Examination revealed a tender mass protruding through the vaginal introitus that was noted to be malodorous. The patient was admitted urgently for stabilization of both an acute kidney injury (AKI) due to urinary retention caused by the obstructing mass and anemia due to the extent of her per vaginal hemorrhage. Surgical management was expedited, and the patient underwent a modified type III radical hysterectomy, bilateral salpingo-oophorectomy, bilateral pelvic lymphadenectomy, and omentectomy. This case required the employment of expert gynecological oncological techniques for optimal patient management. We believe that our identification of such an exceptional case in terms of both the presentation of endometrial carcinosarcoma as NPUI and as an emergency will inform future management of acute presentations of gynecological malignancy.

1 | Introduction

Uterine inversion outside the puerperium is a rare presentation [1]. Reported cases of nonpuerperal uterine inversion (NPUI) are scarce with reviews suggesting that the total number of reported cases is fewer than 200 worldwide [2]. Most documented cases of NPUI are attributed to benign leiomyomas; malignant etiologies are considerably less prevalent accounting for less than 20%–30% [3]. Uterine inversion can manifest as a life-threatening emergency, although this association is primarily observed in obstetric cases. In contrast, NPUI is often believed to develop insidiously [4].

We report here a case of NPUI caused by uterine carcinosarcoma. Notably, this malignancy had been identified only 20 days before the patient's emergency admission. Our case is unique, not only for being one of the rare instances where NPUI is linked to uterine carcinosarcoma but also for being an acute case highlighting the importance of expeditious management of NPUI. For this case, the patient's consent was sought.

Key Clinical Message

NPUI is a severe and life-threatening condition that should always be considered a differential diagnosis when patients present acutely with a background of known uterine malignancy.

2 | Case History and Examination

We present the case of a 73-year-old nulliparous woman who attended her general practitioner with a change in vaginal discharge, fatigue, and poor appetite. She had a history of vitreous detachment and no previous surgery. She had a drug history of paracetamol and no known allergies. Her body mass index (BMI) was stable at 26, she was a nonsmoker and had a performance status of zero.

At first presentation, the patient was unable to tolerate speculum examination; therefore, a pelvic ultrasound was performed showing a thickened endometrium to 5 cm. An urgent referral was made to gynecology for examination under anesthesia and diagnostic hysteroscopy, during which a cavity-distorting polyp was identified.

3 | Diagnosis and Investigation

The histopathology report from the polyp confirmed the presence of uterine carcinosarcoma. Clinical examination by the gynecological oncology team noted a bulky uterus with difficulty visualizing the cervix due to blood in the vagina. No disease was observed in the vagina. Computed tomography (CT) scan of the chest, abdomen, and pelvis with contrast reported a large uterine tumor expanding the endometrial cavity to an anterior posterior (AP) dimension of 78 mm extending to the external os. No extra-uterine disease was noted.

The patient was discussed at the gynecological oncology multidisciplinary team meeting (MDT). As no distant disease was seen on imaging, it was recommended that a total abdominal hysterectomy, bilateral salpingo-oophorectomy (TAHBSO), bilateral pelvic lymph node dissection, and omentectomy should be completed.

Twenty days after initial diagnosis, prior to planned surgical intervention, the patient experienced an exacerbation of symptoms and presented to the emergency department experiencing severe nausea and vomiting. The quantity of vaginal bleeding had significantly increased and was malodorous. Furthermore, she stated that she had not been able to urinate. Vaginal examination revealed an exquisitely tender protruding mass through the introitus with an offensive odor. The mass was noted to be dusky and edematous in nature; no formal measurements were taken at this point due to the severity of pain experienced by the patient. Laboratory blood testing reported a hemoglobin level of 84 g/L (normal range 120–150 g/L) and creatinine levels of 122 μmol/L (normal range 49–90 μmol/L). A bladder scan confirmed that the patient was in urinary retention with 1 L of urine in the bladder. The patient was admitted, and a Foley's catheter was inserted with the aid of a Sim's speculum to relieve the post-renal obstructive acute kidney injury. Intravenous fluids, 2 units of red blood cells, tranexamic acid, and ondansetron were administered. In view of the exacerbation of her symptoms, surgical management was expedited to the same day.

Prior to surgical exploration, several differential diagnoses were considered to account for this acute presentation, including significant tumor spread on the background of a recent diagnosis of a highly aggressive endometrial carcinosarcoma. Bowel involvement and potential bowel obstruction were also investigated due to the patient's clinical presentation of nausea and vomiting. CT (Figure 1) demonstrated a markedly expanded endometrial cavity extending to the external os, measuring up to 76 mm in AP thickness and 140 mm in maximum length. Toward the fundus there is heterogeneously enhancing soft tissue, with no evidence of myometrial invasion or extrauterine disease. The ovaries appear normal. There is no abdominopelvic lymph node enlargement or distant metastatic disease. NPUI was not considered a top differential at this stage with no clinical or radiological evidence of such a rare phenomenon.

4 | Treatment

In theater, the patient was placed in the modified Lloyd-Davies position with the peri-operative risk reduction protocol implemented to reduce the risk of 'Well-leg' compartment syndrome [5], after which a diagnostic laparoscopy was performed for full surgical staging including the presence of peritoneal carcinomatosis that would have required multivisceral resection. Laparoscopy showed no evidence of bowel disease or upper abdominal disease; however, it did confirm uterine inversion due to the gross anatomical distortion and the degree of invagination (Figure 2). No peritoneal disease was noted on laparoscopy.

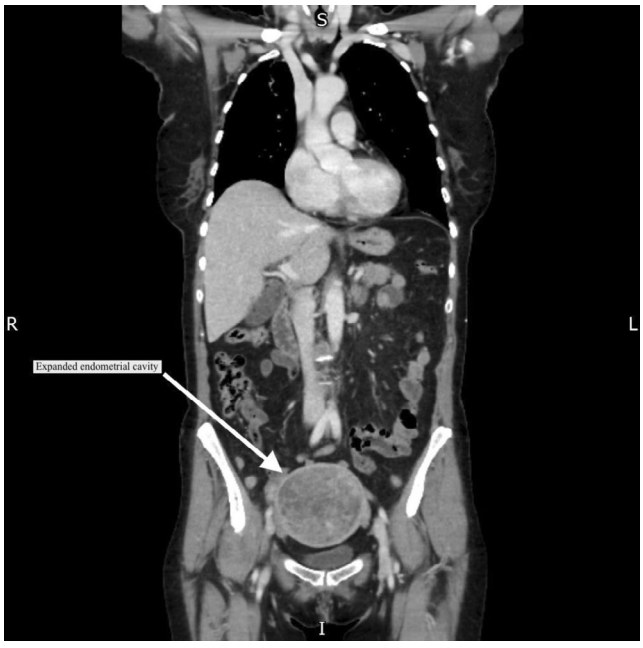


FIGURE 1 | Pre-operative CTAP demonstrating a markedly expanded endometrium extending to the external os.

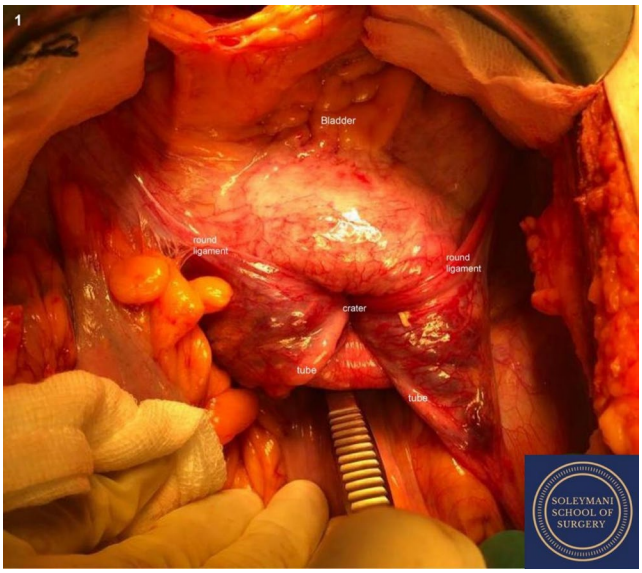
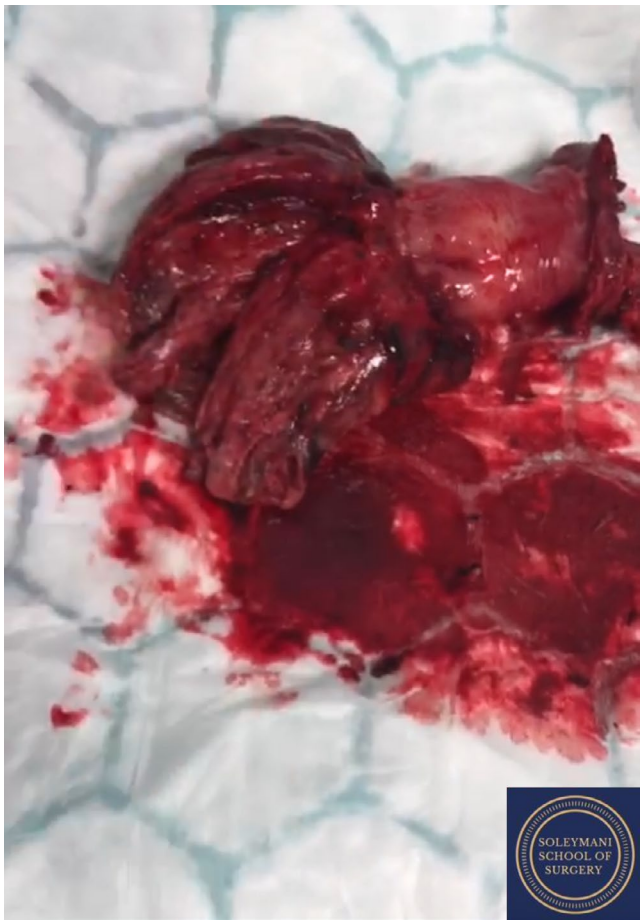


FIGURE 2 | Intra-abdominal view of the pelvis demonstrating the descending round ligaments and fallopian tubes.

Due to the strong suspicion of cervical involvement, at both EUA and laparotomy, a radical hysterectomy was performed in order to obtain clear margins including resection of parametrium for oncological adequacy. Following midline laparotomy and opening of the abdominal wall in layers, the large bowel was mobilized along the Todd lines to the splenic and hepatic flexures. The pelvic sidewalls were opened bilaterally and after visualization of the internal and external iliac vessels, the internal iliac vessel was slung. The ureter was also dissected and slung. The infundibulo-pelvic ligament and round ligament were both ligated. A modified radical hysterectomy was performed using retrograde Hudson's procedure for en bloc total pelvic peritonectomy for optimal cytoreduction with the intention to achieve no residual disease also known as R0. Bilateral pelvic lymphadenectomy and omentectomy were also performed after the spaces had been opened for hysterectomy. The procedure was highly technically challenging due to the presence of a constriction ring; furthermore, the degree of distortion of anatomy made confirmation of clear margins difficult. No repositioning of the uterus was undertaken prior to hysterectomy due to the degree of anatomical distortion. The diagnosis of NPUI was further confirmed in theater upon examination of the specimen in which attempts to restore the uterine inversion were unsuccessful (Video 1). After the specimen appeared to have been completely excised, it was sent for histopathological review. It weighed 665 g in theater. A Robinson's pelvic drain was left in situ at the left iliac fossa. The patient received an intraoperative blood transfusion of 1 unit of red cells and had an estimated blood loss of 300 mL. She was moved to the high dependency unit postoperatively to start her recovery. The patient was discharged on day 12 postoperatively.

5 | Outcome and Follow-Up

The histopathology report described the macroscopic appearance of the tumor emerging from the fundus as necrotic and poorly preserved (Figure 3). It was not possible to determine orientation, and



VIDEO 1 | Video showing inversion of the uterus by the fundal mass. Video content can be viewed at https://onlinelibrary.wiley.com/doi/10.1002/ccr3.71917.

the ovaries and fallopian tubes were not recognizable due to the necrotic nature of the tumor (Video 1). The uterus and cervix approximately measured 80×50×40 mm; however, the tissue mass extended to 150×110×30 mm, suggesting a significant degree of tissue inflammation and edema within surrounding tissues. The endometrium was thickened up to 15 mm, and the myometrium was thickened to 30 mm. Microscopically, the tumor was determined to be an FIGO stage 3a carcinosarcoma (FIGO 2009 Criteria) with no spread to the lymph nodes or omentum. The carcinomatous component appeared to be serous, and the sarcomatous component was homologous. She was referred to clinical oncology and ultimately only received three cycles of carboplatin and paclitaxel, which was ceased due to developing toxicity to these drugs. After several discussions regarding alternative chemotherapy agents, the patient decided against further medical oncological treatment due to her age and the potential side effects. Furthermore, the patient was counseled extensively regarding the role of radiotherapy in endometrial cancer management including the fact it was used to improve postoperative loco-regional control but not her overall survival; however, she was reluctant to undergo further intervention after chemotherapy and decided against this treatment [6]. She remains alive and well more than 5 years after initial treatment and was recently discharged from gynae-oncology after her final CT report did not show any new disease. It is thought that her favorable outcome was positively prognosticated by intraoperatively achieving R0 excision margins

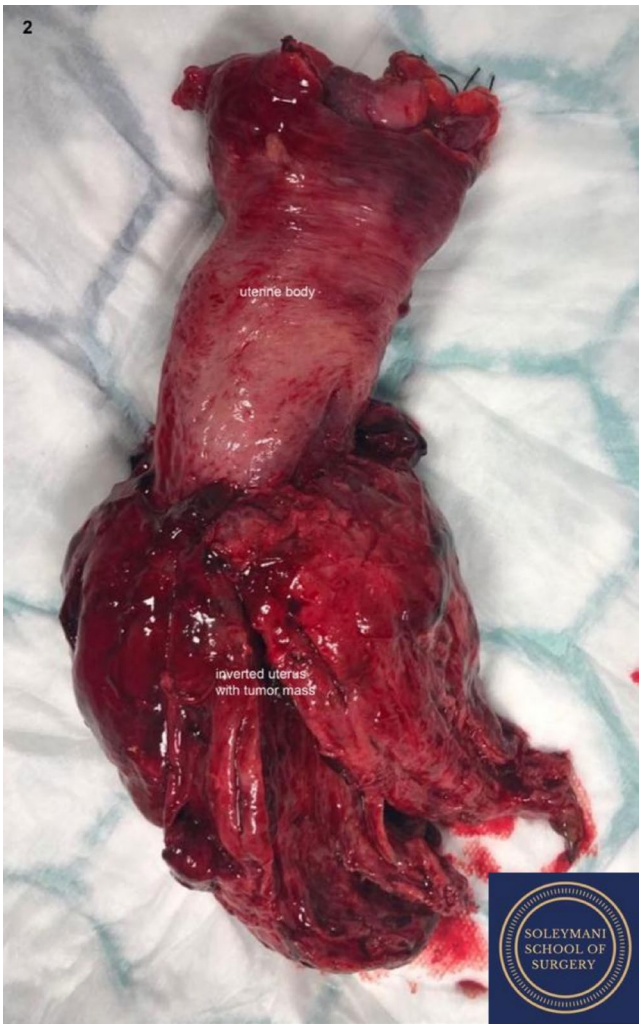


FIGURE 3 | Specimen following modified radical hysterectomy including visualization of the uterine body with the inversion of the uterus and tumor mass.

and the histology of the final specimen reporting a lack of lymphovascular space invasion.

6 | Discussion

Uterine inversion can be defined as invagination of the uterine fundus through the cervix. It is a rare condition and can be classified into either puerperal (obstetric) or nonpuerperal (gynecological) [7]. Most reported cases of inversion are acute obstetric emergencies following delivery resulting in major postpartum hemorrhage with a reported incidence of 1 in 30,000 deliveries [8]; however, due to the even smaller incidence of nonpuerperal hemorrhage, it is difficult to predict the incidence in comparison.

NPUI is usually caused by fundal tumors which pull the uterine fundus into the uterine cavity simultaneously inverting the uterus. Most accounts of NPUI were benign in origin, most reported to be leiomyomas [3]. A search of the literature from 2010 to 2025 returned only 35 cases of malignancy associated with NPUI, of which nine were of the carcinosarcoma (or mixed Müllerian tumor) subtype (Table 1).

The mechanism by which the uterus becomes inverted in nonpuerperal cases is debated, unlike puerperal inversion [44]. It is thought to be a multifactorial process caused by weakening of the uterine wall by the tumor, the weight of the tumor and the attachment site of the tumor [33, 45]. Furthermore, Herath et al. [2] suggested that distension of the myometrium by the tumor may cause expulsive contractions resulting in dilatation of the cervix and subsequent expulsion of the tumor.

As many cases of NPUI are benign, a number of uterine restoration techniques to reposition the uterus have been attempted to preserve fertility. There are both transabdominal and transvaginal methods available including Huntington's, Haultain's,

TABLE 1 | Summary of published case reports of non-puerperal uterine inversion (NPUI) from 2010 to 2025, with documented etiologies.

Associated tumor	Number of cases	Authors
Carcinosarcoma	9	Massinde et al. [9], Mehra et al. [10], Sardar et al. [11], Zhao et al. [12], Kean et al. [13], Misuari-Alihuddin et al. [14], Eouani et al. [15], Khoiwal et al. [16], Azhar et al. [17]
Adenosarcoma	8	Occhionero et al. [18], Salameh et al. [19], Shunmugam et al. [20], Belghith et al. [21], Witt et al. [22], Indraprasta et al. [23], Vardar et al. [24], Ghazizadeh et al. [25]
Rhabdomyosarcoma	6	Ambreen et al. [26], Peng et al. [27], Li et al. [28], Asmouki et al. [29], Sali et al. [30] Rodriguez et al. [31]
Adenocarcinoma	2	Kim et al. [32], Wang et al. [33]
Leiomyosarcoma	2	Free et al. [34], Lghamour et al. [35]
Endometrial stromal sarcoma	2	Sharma et al. [36], Sims et al. [37]
Endometrial squamous cell carcinoma	2	Girija et al. [38], Girish et al. [39]
Undifferentiated sarcoma	1	Singh et al. [40]
Unspecified	3	Tuckett et al. [41], Khorshid et al. [42], Robati et al. [43]

and Spinelli's procedures [46]. However, Herath et al. have suggested there is little efficacy of these procedures, with only 15% of patients being successfully treated. Uomoto et al. [47] highlighted the complex nature of repositioning procedures and the risk of uterine perforation in their case report. Therefore, the mainstay of treatment remains a total hysterectomy in both benign and malignant cases.

Our case details two findings of unusual interest. First, the rare cause of uterine inversion secondary to the presence of a high grade endometrial carcinosarcoma, a high-grade tumor with significant prognostic uncertainty [48, 49]. Second, an acute presentation with anemia and hypovolaemia and an AKI due to urinary retention, which required immediate management and indeed expedited her definitive surgical intervention. Thus, our case highlights the fact that NPUI should be seen as a gynecological emergency much like its obstetric equivalent.

Author Contributions

Aman Kaur More: project administration, writing – original draft, writing – review and editing. **Sarah Louise Smyth:** supervision, writing – review and editing. **Ana Sofia Cerdeira:** writing – review and editing. **Hooman Soleymani majd:** conceptualization, data curation, investigation, supervision, writing – review and editing.

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The authors have nothing to report.

Consent

Patient consent was sought prior to publishing this case report.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

This article does not report the generation of any new datasets beyond the clinical information presented in the case description. All patient data relevant to the case are contained within the article. The literature reviewed in the Discussion: Section 6 is available from the published sources cited in the References. No additional data are available.

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