

Non-puerperal uterine inversion associated with adenocarcinoma of the uterus: A case report

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ABSTRACT

Introduction: Uterine inversion is an uncommon condition characterized by the invagination of the fundus of the uterus through the vagina and is extremely rare in non-pregnancy settings. Non-puerperal uterine inversion is usually precipitated by tumours exerting traction force on the fundus of the uterus, turning the uterus partially or completely inside out. It is most frequently associated with benign tumours such as submucosal leiomyomas; however, malignant tumours are a rare association.

Case Presentation: A 67-year-old woman, G18P18, presented to the emergency department with a bleeding mass that had acutely prolapsed out of the vagina. She had a two-year history of postmenopausal bleeding but had not sought medical advice. She underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy. Pathological evaluation revealed an adenocarcinoma of the uterine fundus, measuring 6 cm in its largest diameter, which invaded the myometrium only superficially. The patient recovered well from the operation with no complications and was referred to an oncologist for further treatment. A computerized tomography scan with intravenous contrast showed no evidence of metastasis.

Conclusion: Uterine sarcoma is a malignant tumour of the uterus that typically presents with vaginal bleeding, and rarely as prolapsed uterine inversion. Uterine inversion rarely occurs outside the puerperal setting; however, when it does occur, the possibility of an underlying malignancy should not be neglected.

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1. Introduction

Non-puerperal uterine inversion is a rare condition in which the internal surface of the uterus protrudes through the vagina. This type of inversion is typically associated with a pathological process and submucosal leiomyomas are considered the most frequent gynaecological cause. Endometrial polyps, uterine sarcomas, endometrial carcinomas and mixed mesodermal tumours are also potential causes. Uterine inversion can be acute or chronic. Acute manifestation presents with sudden expulsion of the uterus accompanied by pain and haemorrhage, while chronic manifestation presents with pelvic discomfort and irregular uterine bleeding until the uterus eventually protrudes through or out of the vagina. Management is surgical and largely depends on histological results.

2. Case Presentation

A 67-year-old woman, G18P18, presented to the emergency department after an acute expulsion of a mass out of the vagina that was associated with pain and bleeding (Fig. 1). She had tachycardia (110 bpm) and her blood pressure was 160/90 mmHg; however, all other vital signs were stable and within normal limits. She had a two-year history of postmenopausal bleeding but she had not sought medical advice. She denied any prior episodes of uterine prolapse. On initial examination, the patient had a mass protruding from her vagina with moderate bleeding from the distal portion of the mass, along with multiple tears and loss of tissue. A pelvic ultrasound in the emergency department showed an absence of the genital organs, with no free fluid in the pelvis. A diagnosis of non-puerperal uterine inversion with prolapse was made and she was transferred to the operating room to undergo an abdominal hysterectomy with bilateral salpingo-oophorectomy.

On laparotomy, her pelvic anatomy was seen to be distorted. The uterus was completely inverted, with both adnexal structures incarcerated and covered within the inverted fundal cup. Attempts to reposition the uterus abdominally by grasping both round ligaments using

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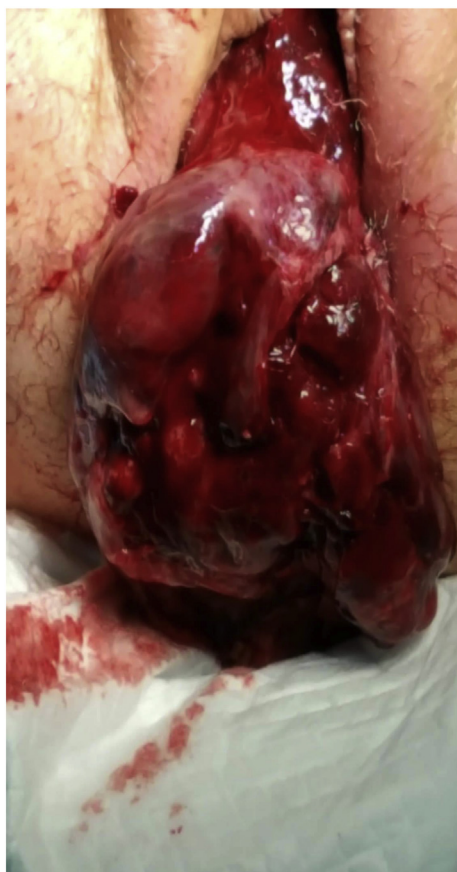


Fig. 1. The prolapsed uterus.

Huntington's and Haultain's method failed. The ureters were grossly dilated and distorted; therefore, a urologist placed bilateral ureteric stents to prevent any damage to the ureters. Hysterectomy with bilateral salpingo-oophorectomy was performed. The patient recovered well from the operation, with no complications.

On pathological examination, the tumour was located in the uterine fundus. The specimen showed a polypoid adenocarcinoma of the uterus measuring 6 cm in its largest diameter, with no involvement of the adnexal structures. It invaded the myometrium superficially, with no evidence of lymphovascular invasion. The remaining endometrium showed atrophic changes without evidence of malignancy. The ovaries were both normal. A computerized tomography (CT) scan with intravenous contrast was recommended by the oncologist. Imaging showed no evidence of metastasis and the final staging was pT1bcNOM0. As per the guidelines for this stage, a 6-month repeat CT scan with intravenous contrast was planned.

3. Discussion

Uterine inversion is an uncommon condition characterized by the invagination of the uterine fundus through the uterine cavity, reaching the cervix or beyond the cervix. Uterine inversion can be puerperal or non-puerperal. The puerperal type is a life-threatening emergency that occurs in the third stage of labour; it has a 15% mortality rate due to bleeding and shock [1,2]. The non-puerperal type is mainly associated with uterine tumours located at the uterine fundus, which force the fundus to invert into the uterine cavity. It has been reported that the majority of the cases in the literature were associated with benign tumours of the uterus, mainly leiomyomas [3].

Uterine sarcoma is a rare group of heterogeneous tumours of the uterus with variable pathology and natural history [4,5]. The incidence of uterine sarcoma is 3–7% of all malignant tumours of the uterus, and

it occurs mostly in women who are over 50 years old [6]. Uterine sarcomas include leiomyosarcoma, endometrial stromal sarcoma, adenocarcinoma, fibrosarcoma and undifferentiated uterine sarcoma [4]. Uterine sarcomas present with vaginal bleeding, lower abdominal pain and abdominal distension, and can rarely be complicated by non-puerperal uterine inversion. A search of the literature from 1887 to 2019 returned 49 cases of uterine sarcoma associated with uterine inversion. It is worth mentioning that carcinosarcomas, also known as “mixed müllerian tumours”, are tumours of epithelial rather than mesenchymal origin and are no longer considered to be uterine sarcomas [4], which decreases the number of reported uterine sarcomas associated with non-puerperal uterine inversion to 35 cases (Table 1).

Uterine adenocarcinoma is the rarest subtype, accounting for only 5% of all uterine sarcomas [7]. In common with other uterine sarcomas, it presents mostly with abnormal uterine bleeding. The majority of patients present with stage I disease, with a 5-year survival of 60–80% [7]. Non-puerperal uterine inversion associated with adenocarcinoma is extremely rare, and extensive literature review revealed only one case, reported by Occhionero in 2012 [8] (Table 1). Standard care in the management of early-stage uterine sarcomas consists of total abdominal hysterectomy and bilateral salpingo-oophorectomy. As the risk of lymph node and omental metastases is negligible, it is best to avoid the morbidity of bilateral pelvic lymphadenectomy and omentectomy. In addition, as adenocarcinomas are indolent and low-grade malignancies, cytotoxic chemotherapy is unlikely to be beneficial and there are no studies supporting the role of radiation treatment [4].

4. Conclusion

Uterine sarcoma is a rare malignant tumour of the uterus that typically presents with vaginal bleeding, and rarely as uterine inversion with prolapse. Uterine inversion rarely occurs outside the puerperal period; however, when it does occur, the possibility of an underlying malignancy should not be neglected. It is also important for postmenopausal vaginal bleeding to be thoroughly investigated and patients should be told not to delay seeking medical advice.

Table 1

All reported cases of non-puerperal uterine inversion associated with uterine sarcoma.

Associated tumour	Number of cases	Authors
Unspecified sarcoma	22	Simpson, 1887; Targett, 1897; Thorn, 1911 (4 cases); McCullagh, 1925; Jones, 1951; Salisbury et al., 1958; Lana, 1962; Bernardine and Alessandro, 1963; Ehrlich and Bonaventura, 1977; Holzer, 1979; Bensaid et al., 1996; Lupovitch et al., 2005; Zahra Etekhar et al., 2005; Pandit, 2006; Mechery, 2009; Tuckett et al., 2010; John David Tuckett et al., 2010; Korshid and Al-Badawi, 2011; Sims et al., 2012
Leiomyosarcoma	7	Kelly, 1898; Spencer, 1919; Mwinyoglee et al., 1997; Rattray, 2000; Takano et al., 2001; Buyukkurt et al., 2007; RekhaSachan et al., 2012
Endometrial stromal sarcoma	4	Clarence E. et al., 1977; Louw and Schouwenburg, 1980; Michael G. Conner et al., 2004; Korshid et al., 2011
Adenocarcinoma	1	Occhionero et al., 2012
Fibrosarcoma	1	Reich and Nechtow, 1946
Undifferentiated uterine sarcoma	0	
Malignant mixed Müllerian sarcoma	14	Craig, 1958; Zorn et al., 1990; Mwinyoglee et al., 1997; Takano et al., 2001; Tjalma, 2003; M. Moodley & J. Moodley, 2003; Hanprasertpong, 2004; GennaroCormio et al., 2005; Cormio, 2006; Gemer, 2007; Sinha et al., 2007; Anthony N.Massinde et al., 2012; Mehra et al., 2013; Zhao et al., 2018
Total cases	49	

Reference details are available from the corresponding author.

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Alaa Eddin K. Salameh wrote first draft of the paper and revised the manuscript.

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Raha M. Almarzooqi wrote first draft of the paper and revised the manuscript.

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Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent

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Provenance and peer review

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