



Uterine adenosarcoma presenting as uterine inversion: A case study

Caitlin Witt^{a,*}, Chelsey Vranes^b, Leslie H. Clark^b

^a Louisiana State University Health Sciences Center, Baton Rouge, LA, USA

^b UNC Department of Gynecologic Oncology, Chapel Hill, NC, USA

ARTICLE INFO

Keywords:

Adenosarcoma
Uterine Inversion
Non-Puerperal
Inversion

1. Introduction

Non-puerperal uterine inversion is a rare complication where the fundus of the uterus turns inside out and prolapses through the cervix [1]. The incidence of non-pleural uterine inversion is unknown and is most commonly caused by benign leiomyomas, while it is rarely associated with uterine sarcomas [1,2]. Uterine adenosarcoma is a biphasic tumor composed of benign epithelium and malignant stroma that accounts for 5–10% of uterine sarcomas [1,2]. It is hypothesized that rapid growth of tumor and softening of uterine wall due to tumor enlargement can cause the rare complication of uterine inversion [1].

2. Case description

A 35-year-old G1P0010 female presented with pelvic mass and acute blood loss anemia. The patient had no significant past medical history and past surgical history of dilation and curettage after spontaneous abortion. Laboratory findings on admission notable for a hemoglobin of 5.1 ultimately requiring a total of six units of packed red blood cells prior to being transferred to our tertiary care center. Exam under anesthesia with biopsy was also performed prior to transfer with a large mass protruding from and encompassing the cervix with active bleeding. Computed Tomography scan of abdomen and pelvis was notable for distended vagina containing fluid and gas with abnormal configuration of uterus and apparent uterine inversion into the vagina. Magnetic Resonance Imaging (MRI) of the Pelvis confirmed the uterus to be inverted with associated distension of the vaginal canal and the left ovary with partial intussusception within the inverted uterus [Fig. 1].

The patient was then transferred for higher level of care and

gynecologic oncology consultation. Decision made to proceed with emergent surgery due to continued bleeding, ongoing blood transfusion requirements, and concern for superimposed infection. A total

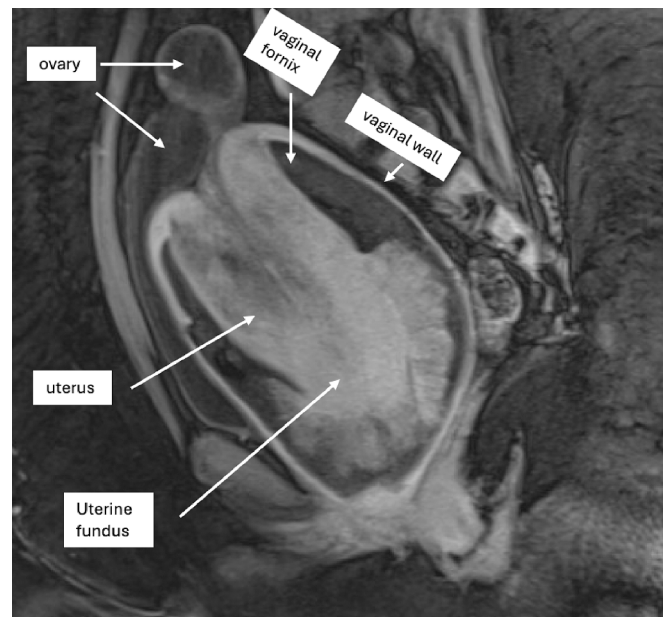


Fig. 1. MRI pelvis.

* Corresponding author at: 500 Rue De La Vie Suite 404, Baton Rouge, LA 70817, USA.

E-mail address: cwitt@lsuhsc.edu (C. Witt).

<https://doi.org/10.1016/j.gore.2024.101398>

Received 17 March 2024; Received in revised form 11 April 2024; Accepted 15 April 2024

Available online 16 April 2024

2352-5789/© 2024 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).



Fig. 2. Endometrial adenocarcinoma non-pleural uterine inversion.

abdominal hysterectomy with bilateral salpingectomy was performed. On bimanual exam a $5 \times 8 \times 8$ cm exophytic mass within the vagina was noted with no evidence of invasion into vaginal tissue or bilateral sidewalls. Intra-abdominal findings were notable for complete uterine inversion with distorted fundal contour and ovaries protruding cephalad from inverted uterine fundus. No evidence of extrauterine pelvic disease noted at time of surgery. An unsuccessful attempt was made to reduce uterus to proper anatomic location. Given altered anatomy, an anterior colpotomy was created using handheld malleable to delineate anterior vaginal fornix, and a retrograde hysterectomy was performed [gross specimen shown in Fig. 2]. Frozen pathology showed extensive necrosis and concern for sarcomatous growth, therefore no lymph node

procedures were performed given normal nodal evaluation on preop MRI. The ovaries were left in situ given normal appearance, presumed sarcoma, and the patient's young age.

Final histopathologic examination revealed a FIGO Stage IB endometrial adenocarcinoma with sarcomatous overgrowth and heterologous rhabdomyosarcoma. Acute inflammation and necrosis involving tumor surface and extensive edema within the tumor was noted. The mass was composed of benign endometrial glands and malignant stroma composed of cells with spindled nuclei arranged in sheets and nests [Fig. 3]. Other areas of stromal cells showed features of strap cells with cytoplasmic striation and cells with eccentric nuclei and bright eosinophilic cytoplasm. CD10 positive stain noted in some *peri-glandular*

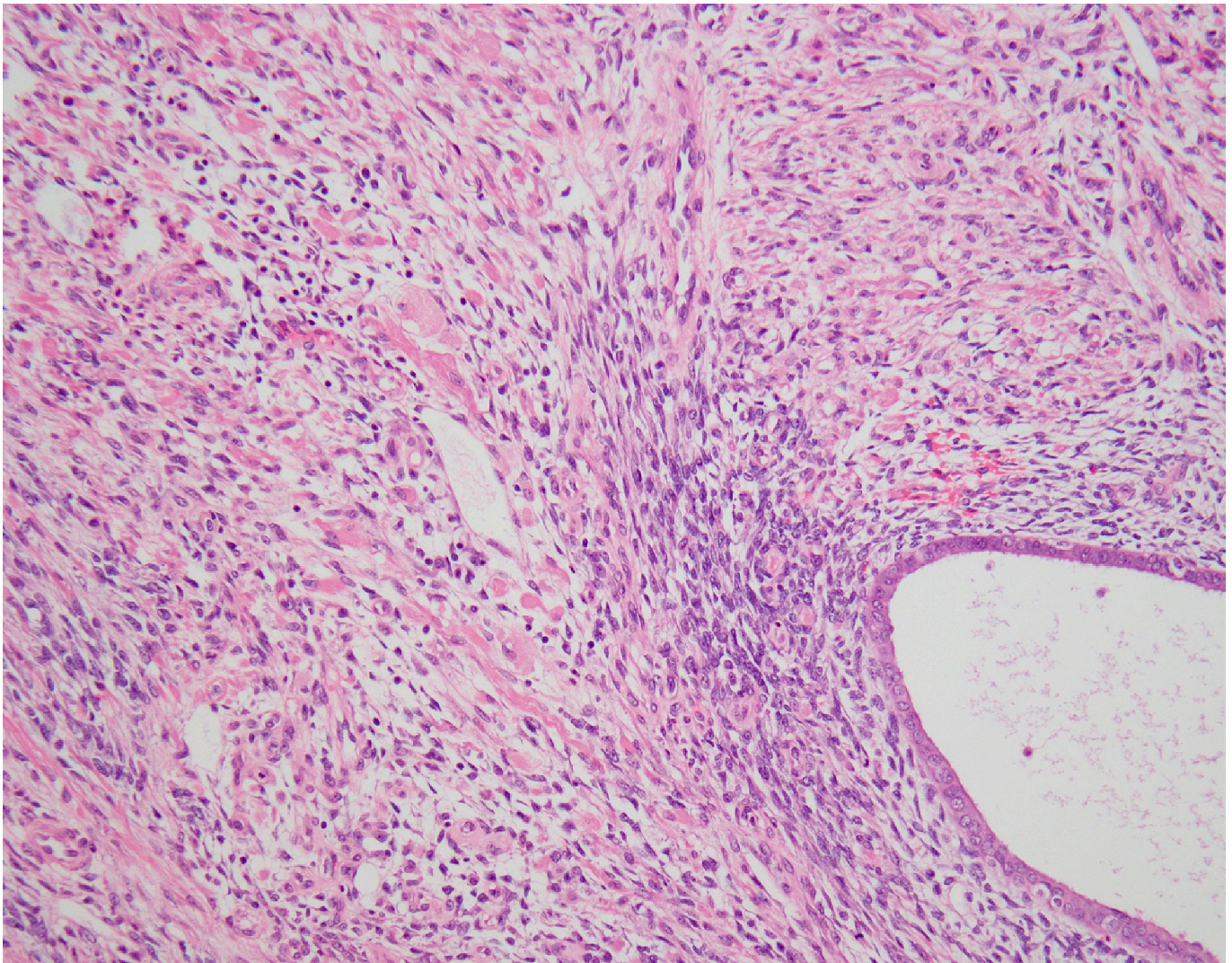


Fig. 3. Endometrial adenocarcinoma pathology.

stroma and desmin/myogenin noted positive in many of the malignant stromal cells. Estrogen receptor was 20 % positive and progesterone receptor was 30 % positive. Myometrial invasion was present to the depth of 12 mm and lymphovascular invasion was negative. All margins were noted to be negative for sarcoma.

3. Discussion

Surgical management is the mainstay of treatment for non-puerperal uterine inversion once the patient is stabilized; if malignancy is suspected, then a hysterectomy should be performed [2]. The diagnosis of uterine inversion is often made with imaging, with MRI thought to be the most sensitive for diagnosis. However, if the diagnosis is unclear, laparoscopy can be used to confirm inversion [1,2]. As demonstrated in this case report, patients with non-puerperal uterine inversion can present in unstable condition with significant risk for bleeding and infection. Adenosarcoma most commonly presents with symptoms of abnormal vaginal bleeding [3,4]. However, more uncommon presentations include uterine enlargement, pelvic mass and protruding cervical tumors [4].

When non-puerperal uterine inversion is encountered attempt to reduce uterus to obtain normal anatomy can be performed. As demonstrated in this case when normal anatomy cannot be restored retrograde

abdominal hysterectomy can be utilized as a surgical technique. Retrograde abdominal hysterectomy is a technique that can be used to perform hysterectomy when cervicovaginal junction is not clearly identified or a large vaginal tumor is present [5]. This technique involves initial bladder mobilization and colpotomy creation at the vesicouterine junction [5]. The uterine arteries and cardinal ligament are then clamped and divided at the level of the internal cervical os [5]. The incision at the anterior vaginal wall is extended and the bilateral uterosacral ligaments are ligated [5]. A retrograde hysterectomy is performed without visualization of the ureters. Ureter injury is avoided by close contact with the portio vaginalis uteri when transecting the parametrium and uterosacral ligaments [5].

4. Conclusion

This case report demonstrates a rare presentation of an adenosarcoma in a pre-menopausal person presenting with a similarly rare uterine inversion who ultimately required an abdominal hysterectomy due ongoing vaginal bleeding, instability, and high suspicion of malignancy.

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

CRediT authorship contribution statement

Caitlin Witt: . **Chelsey Vranes**: Writing – review & editing, Writing – original draft, Conceptualization. **Leslie H. Clark**: Writing – review & editing, Supervision, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

- [1] Occhionero, M., Restaino, G., Ciuffreda, M., Carbone, A., Sallustio, G., Ferrandina, G., 2012. Uterine inversion in association with uterine sarcoma: A case report with MRI findings and review of the literature. *Gynecol Obstet Invest* 73 (3), 260–264. <https://doi.org/10.1159/000334311>.
- [2] Herath, R.P., Patabendige, M., Rashid, M., Wijesinghe, P.S., 2020. Nonpuerperal Uterine Inversion: What the Gynaecologists Need to Know? *Obstet Gynecol Int.* <https://doi.org/10.1155/2020/8625186>.
- [3] Pinto, A., Howitt, B., 2016. Uterine Adenosarcoma. *Arch Pathol Lab Med.* 140 (3), 286–290. <https://doi.org/10.5858/arpa.2014-0523-RS>.
- [4] Wang, Q., Sun, S., Cai, J., Yang, L., Lv, G., Yang, Q., 2023. Uterine Adenosarcoma: A Case Report and Review of the Literature 13.
- [5] Hiramatsu, Y., 2019. Retrograde abdominal hysterectomy. *Surg. J.* 05 (S 01), S27–S32. <https://doi.org/10.1055/s-0039-1683919>.