CASE REPORT

Uterocutaneous fistula as the primary presentation of a gynaecological malignancy

Liesel Elisabeth Hardy,¹ Yee Leung^{1,2}

¹Department of Gynaecological Oncology, King Edward Memorial Hospital for Women, Perth, Western Australia, Australia

²School of Women's and Infants' Health, University of Western Australia, Perth, Western Australia, Australia

Correspondence to
Dr Liesel Elisabeth Hardy,
lieselhardy@gmail.com

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SUMMARY

We report a case of high-grade gynaecological carcinoma presenting as a uterocutaneous fistula. A 59-year-old woman presented with a discharging abdominal wall wound. Imaging confirmed a large solid pelvic mass forming a sinus tract with the anterior abdominal wall. The tract tunnelled through a previous caesarean section scar. Biopsy indicated a high-grade gynaecological carcinoma, with features suggestive of endometrioid adenocarcinoma. The patient underwent two cycles of chemotherapy. Despite this, the mass increased in size and was complicated by abdominal wall wound breakdown. Chemotherapy was ceased. Surgical and palliative options are under consideration.

BACKGROUND

We report a case of a high-grade gynaecological carcinoma causing a uterocutaneous fistula. A fistula is the abnormal communication between two epithelialised surfaces. Gynaecological fistulas form in the context of surgery (including caesarean section), infection, trauma, radiation therapy and malignancy of the gynaecological tract or surrounding pelvic organs.¹⁻⁴ The most common fistulas associated with gynaecological malignancy are vesicovaginal and enterovaginal.⁵ Uterovesical fistulas are not uncommon. However, uterocutaneous fistula formation is a rare phenomenon, with only a handful of case reports having been published. Furthermore, uterocutaneous fistulas are known to develop as a result of radiation therapy, surgical or postpartum complications.⁶ To our knowledge, there are no prior case reports of a uterocutaneous fistula as the primary presentation of a uterine malignancy.

CASE PRESENTATION

A 59-year-old woman presented to the emergency department of a regional hospital reporting a 1-week history of a tender discharging anterior abdominal wall wound. The wound was at the site of a previous midline caesarean section scar. The patient had also noticed an abdominal mass on palpation, had unintentionally lost 30 kg of weight in 12 months, experienced reduced appetite, mild constipation and intermittent per-vaginal spotting. The patient had no significant previous medical history and was not on any regular medications. Obstetric history was significant for two caesarean sections over 20 years prior. Gynaecological history was significant for

tubal ligation. The patient reports one abnormal Pap smear at age 22 followed by a cone biopsy and no further intervention. Her last Pap smear 10 years prior was normal. She underwent menopause at 49 years of age and had never been on Hormone replacement therapy (HRT). She was a current smoker of 20 cigarettes per day and had been for 40 years. She drank two glasses of wine per day. The only family history was of paternal bowel cancer at age 85.

On examination, the patient had a palpable central mass arising from the pelvis extending suprapubically, approximately 10 cm in diameter. The abdomen to the level of the umbilicus was firm to palpation. An abdominal wall wound (5×3 cm) was situated midway along a midline caesarean scar and contained pus. Examination of external genitalia was normal and speculum examination was normal. On bimanual examination, the uterus was not appreciably separate to the mass. Rectal examination was normal. Breast examination was normal, no supraclavicular fossa, axillary or groin lymphadenopathy.

INVESTIGATIONS

Bloods and microbiology

Initial presentation (28 July 2017)

Cancer antigen (CA) 125 78. Carcinoembryonic antigen (CEA) 6. Alpha-fetoprotein (AFP) 3. CA 19.9 3. Lactate dehydrogenase (LDH) 256. White cell count: $18.04 \times 10^9/L$. Platelet count: $515 \times 10^9/L$. Neutrophils $15.23 \times 10^9/L$. C reactive protein 78. All other results of Urea, electrolytes, creatinine (UEC), Full blood examination (FBE) and liver function tests (LFTs) were normal.

Wound swab: growth of *Escherichia coli*.

Urine: no growth.

Second admission (24 September 2017)

Wound swab: mixed coliforms, mixed skin flora, few gram-positive cocci and few gram-negative cocci on microscopy.

Vaginal swabs: moderate gram-positive cocci, a few mixed bacteria, moderate leukocytes, few lactobacilli.

Imaging

Pelvis ultrasound (1 August 2017)

There is a solid mass (77×63×99 mm) within the pelvis, and the site of origin is uncertain; a uterine cause would have to be considered. The ovaries have not been seen separate to this mass.

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Figure 1 A sagittal section from the initial CT scan. Demonstrating a heterogeneous pelvic mass arising from the expected location of the uterus. The mass erodes into the inferior abdominal wall via a sinus tract.

CT abdomen (1 August 2017)

1. The 120 mm heterogeneous pelvic mass arises from the expected location of the uterus and cervix and is most likely uterine in origin. Appearances are felt most likely secondary to an aggressive uterine malignancy, including uterine sarcoma. A complex pelvic abscess could also give these appearances, although this is considered less likely in view of the patient's afebrile state.
2. There is erosion of the mass into the inferior abdominal wall and overlying soft tissues via a sinus tract, and suspected fistula formation with the urinary bladder.
3. There is loss of the fat planes between the mass and the sigmoid colon and transverse colon without definite evidence of fistulation at these sites.
4. Left para-aortic lymphadenopathy.

CT chest (4 August 2017)

No evidence of thoracic metastatic disease.

Figure 2 A coronal section from a CT scan at initial presentation, demonstrating the pelvic mass.

CT cystogram (9 August 2017)

Contrast is noted within the bladder and there are multiple small locules of gas within the bladder. However, there is no contrast extravasation. No vesicouterine fistula is demonstrated.

PET scan (23 August 2017)

1. The large pelvic mass is intensely FDG avid and would be in keeping with a primary malignancy. This has increased in size since the recent CT.
2. Inflammatory activity within the lower abdominal wall fistula.
3. Moderate-grade focal uptake in an apparent soft-tissue nodule anterior to the left adrenal is suspicious for a metastasis.
4. Low-grade activity in an enlarged left para-aortic node is also suspicious for a metastasis.
5. No evidence of extra-abdominal metastatic disease.

CT abdomen/pelvis (24 September 2017)

The mass in the pelvis (10.6.10.7×13.1 cm) has increased in size with multiple air pockets communicating to the anterior abdominal wall, possible mass abscess. Small hypodense lesion in the right labia may also represent a small abscess.

MRI pelvis (23 November 2017)

Appearances of a similar sized pelvic mass communicating to the anterior abdominal wall. Either oedema or peritoneal disease is now present within the flanks, although it is noted that there was no FDG uptake on the PET study prior.

Figure 3 A sagittal section from CT scanning at second presentation (2 months after initial CT) with abdominal wall wound breakdown. CT demonstrates an increase in the size of the pelvic mass with multiple air pockets communicating to the anterior abdominal wall.

Histology/cytology

Ultrasound-guided biopsy (8 August 2017)

Malignant; high-grade carcinoma of gynaecological origin. Although the appearances are largely undifferentiated, there are some features that favour a high-grade endometrioid adenocarcinoma. High-grade serous carcinoma is not excluded on this core material.

Pap smear (6 August 2017)

Smear mainly shows acute inflammatory exudate with red blood cells. An endocervical component is not present. There are a small number of atypical cells, demonstrating large nuclei and prominent nucleoli. The cells may be uterine in origin but their

Figure 4 A coronal section from CT scanning at second presentation (2 months after initial CT) with abdominal wall wound breakdown. CT demonstrates an increase in the size of the pelvic mass with multiple air pockets.

precise nature is not clear. Negative for human papillomavirus genotypes.

DIFFERENTIAL DIAGNOSIS

Uterine malignancy, actinomycosis, sarcoma, pelvic abscess and ovarian malignancy.

TREATMENT

At initial presentation in July 2017, the patient was admitted to a tertiary hospital. Blood tests showed a neutrophilia and raised inflammatory markers. She was commenced on intravenous antibiotics (ceftriaxone) and analgesia. A pelvic ultrasound showed a pelvic mass 77×63×99 mm of suspected uterine origin. A CT scan (figures 1 and 2) showed a heterogeneous pelvic mass in keeping with an aggressive uterine malignancy. The lesion showed erosion into the inferior abdominal wall and overlying soft tissues via a sinus tract, with suspected fistula formation with the urinary bladder. While there were locules of gas noted in the urinary bladder, a CT cystogram did not identify a vesicouterine fistula. The patient denied any pneumaturia and her urine culture and microscopy were negative for infection. An ultrasound-guided biopsy revealed a high-grade carcinoma of gynaecological origin, with features suggestive of high-grade endometrioid adenocarcinoma.

The patient was discussed at a tumour multidisciplinary meeting and was offered neoadjuvant chemotherapy. She underwent two rounds of chemotherapy (carboplatin and docetaxel) in September 2017. While she tolerated this well,

she presented to a regional hospital emergency department 4 days after her second round of chemotherapy with copious discharge from her abdominal wound. A repeat CT scan (figures 3 and 4) showed that the pelvic mass had enlarged since her scan 7 weeks prior. The mass had gas locules and necrotic material centrally. Necrotic material from within the mass, as well as urine, appeared to have ruptured via the uterocutaneous fistula to the abdominal surface. The patient was hypotensive and febrile with a neutrophilia. She was treated for sepsis and commenced on intravenous antibiotics (amoxicillin, metronidazole, gentamicin). The patient completed her course of antibiotics and improved clinically and biochemically during her admission and was discharged home. However, chemotherapy has been ceased. An MRI in November 2017 demonstrated a thick-walled posterior urinary bladder that appears contiguous with the mass, as well as disease that extends to the pelvic side walls. Surgical options were considered in the gynaecological oncology multidisciplinary team discussion with the sarcoma team. The case was deemed unresectable given the non-curative intent of surgery, extensive excision of the anterior abdominal wall soft tissue that would be required and potential risk for major postoperative complications in an immunocompromised patient.

Learning points

- ▶ Gynaecological malignancies can present as uterocutaneous fistulas.
- ▶ Complications of radiation therapy, surgery and infection are the most common cause of uterocutaneous fistulas.
- ▶ Gynaecological malignancy invading the anterior abdominal wall provides a challenge when surgical management is under consideration.

OUTCOME AND FOLLOW-UP

The patient is having ongoing input from the palliative care physicians to alleviate her symptoms.

DISCUSSION

A uterocutaneous fistula caused by a gynaecological malignancy has not been reported in the literature previously. However, there have been case reports of uterocutaneous fistulas forming as a complication of radiation therapy, surgery (caesarean section) and infection.^{1 3 6}

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