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Endometrial adenocarcinoma arising in a pelvic implant following uterine morcellation: A case report

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ABSTRACT

We report a unique case of a 60-year-old woman developing endometrial cancer in a uterine deposit 18 years after she had undergone laparoscopic subtotal hysterectomy with morcellation for benign pathology. She had used unopposed estrogen as menopausal hormone therapy. She presented with a pelvic mass that was causing pressure symptoms. On imaging, the mass had an enhancing vascular nodular component and appeared to abut normal ovaries and the residual cervix. She proceeded to laparotomy, where a 12 cm pelvic mass was found morbidly adherent to the bladder anteriorly and to the cervical stump. The pelvic mass was excised, and trachelectomy and bilateral salpingo-oophorectomy were performed. Adjacent to this mass was a separate, 5 cm adnexal mass, which was also excised. Histopathology of the smaller pelvic mass was consistent with endometrial adenocarcinoma grade 1, arising in complex endometrial hyperplasia with atypia surrounded by myometrium consistent with a uterine implant. This case highlights the need for consideration and discussion of possible risks of subtotal hysterectomy and morcellation of the uterus for benign disease. Furthermore, given the results in this patient, the use of unopposed estrogen in such patients is discouraged due to possible effects on any residual endometrium still present.

1. Introduction

The use of morcellation in gynaecological surgery remains controversial. While morcellation has allowed patients with fibroid uteri to undergo minimally invasive surgery, morcellation of an undiagnosed malignancy results in seeding of the malignant cells throughout the pelvis and abdomen [1]. As such, morcellation of any type is contraindicated in women with an established cancer, pre-malignant lesions or a suspected malignancy [1]. Prior to a laparoscopic hysterectomy, where morcellation may be considered, a thorough pre-operative evaluation should be conducted to identify possible malignancy, including appropriate imaging, cervical cancer screening and endometrial tissue sampling [2]. Nevertheless, no imaging modality can definitively rule out the presence of an invasive malignancy, particularly in the setting of presumed uterine fibroids.

Subtotal or supracervical hysterectomy (the removal of the uterus with preservation of the cervix) has been preferred to total hysterectomy due to a perception that it was better able to preserve sexual function. However, further research, including a systematic review, found there was no difference between subtotal and total hysterectomy in either short- or long-term urinary, bowel or sexual function [3]. Subtotal hysterectomy may also be the preferred surgical technique for known benign disease of the uterus when adhesions or previous uterine surgery may cause more surgical morbidity than a total hysterectomy.

The following case is unique in that the patient had no malignancy evident on her initial pathology examination of the uterus following a subtotal hysterectomy, only leiomyoma. However, following treatment with unopposed estrogen for menopause she developed endometrial cancer in a pelvic deposit of uterine tissue.

2. Case Presentation

A 60-year-old woman was referred with an indeterminate pelvic mass. Eighteen years earlier she had undergone subtotal laparoscopic hysterectomy for benign disease. The ovaries and tubes were preserved at the time and final pathology was benign. While no formal operation note was available for review, given the operation was performed 18 years ago, this would presumably have been done without containment in a bag, as was the practice at the time.

On review, her main symptoms were urinary frequency and worsening constipation. The patient had been using menopausal hormone therapy (MHT) in the form of unopposed estrogen a few years after surgery up to the current presentation. She was otherwise well, took no other medications, was a non-smoker and had no other significant medical, family or psychosocial history. On examination there was no

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lymphadenopathy. Pelvic examination was normal, with no palpable masses and a normal cervix, vulva and vagina.

Initial review of imaging of the pelvis with ultrasound and computed tomography (CT) showed a 12 cm pelvic mass possibly associated with the upper vagina. There was no ascites or sign of metastatic disease but her cancer antigen CA-125 was mildly elevated at 54 kU/L (normal 0–35 kU/L).

The patient's case was reviewed at a tumour board meeting. The CT showed the left ovary was adjacent to a pelvic mass containing a solid projection (Fig. 1). It was thought to be either a para-ovarian lesion or part of the remnant cervix. The ultrasound demonstrated the same pelvic lesion, with a solid and vascular nodular enhancing component abutting the left ovary and vaginal vault and cervix (Fig. 2).

The patient proceeded to an abdominal trachelectomy with removal of pelvic mass and bilateral salpingo-oophorectomy as per the recommendations of the tumour board meeting. Intra-operatively, a 12 cm mass was found with a solid and cystic component containing fluid consistent with old blood (Fig. 3). The mass was morbidly adherent to the bladder anteriorly and to the residual cervix, suggesting it may have been a portion of residual lower uterine segment. Separate to this mass was an adnexal mass measuring 5 cm, which did not appear to be arising from the cervical mass and was not clear on pre-operative imaging. The ovaries and fallopian tubes appeared otherwise normal.

Final pathology of the pelvic deposit was consistent with grade 1 endometrioid adenocarcinoma with areas of complex endometrial hyperplasia with atypia within a pelvic myometrial implant (Fig. 4). There was no evidence of myometrial invasion in the mass. The residual cervix was unaffected by tumour but had endometriosis present within the cystic component attached to it. The ovaries and fallopian tubes and residual cervix were benign.

The patient was reviewed by the radiation oncology team post-operatively for discussion regarding the role of adjuvant radiation treatment. As there was no myometrial invasion and negative peritoneal washings, the risk of local recurrence was thought to be fairly low. Furthermore, the tumour was found to be estrogen and progesterone receptor positive, and, given that the patient had ceased her MHT, the risk of recurrence was thought to be even lower. Given the potential side-effects associated with radiotherapy to the pelvis and the low risk of recurrence, both the patient and the radiation oncologist decided to proceed with ongoing observation and reserve radiotherapy for treatment of local recurrence.

The patient will continue with clinical follow-up with the gynaecological oncology team.

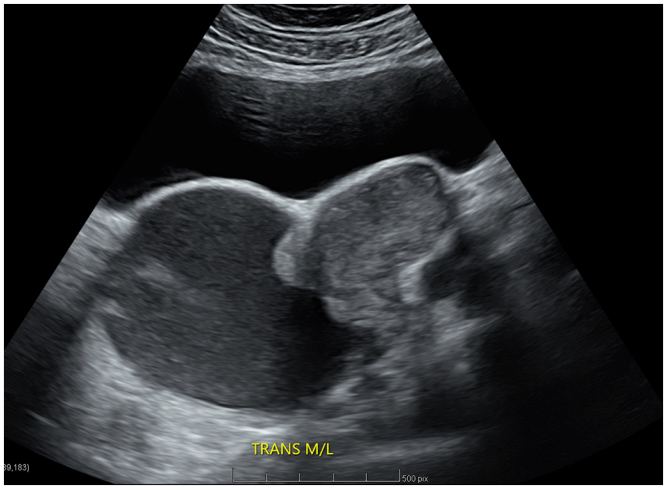

Fig. 2. Ultrasound scan showing solid enhancing nodule adjacent to vagina and remnant cervix.

3. Discussion

As discussed, morcellation of the uterus in suspected or confirmed malignant disease is contraindicated due to the known risk of seeding of the malignancy throughout the abdomen and pelvis [1]. Morcellation of the uterus can also have benign complications, with multiple case reports of benign pelvic implants progressing to atypical hyperplasia, iatrogenic endometriosis, peritoneal adenomyoma and peritoneal leiomyomatosis [1]. While these conditions are not malignant, they can present with pain or mass effect, requiring secondary surgery and increasing morbidity [4,5]. This case appears to be unique, as a search of the literature found no other cases of endometrioid adenocarcinoma

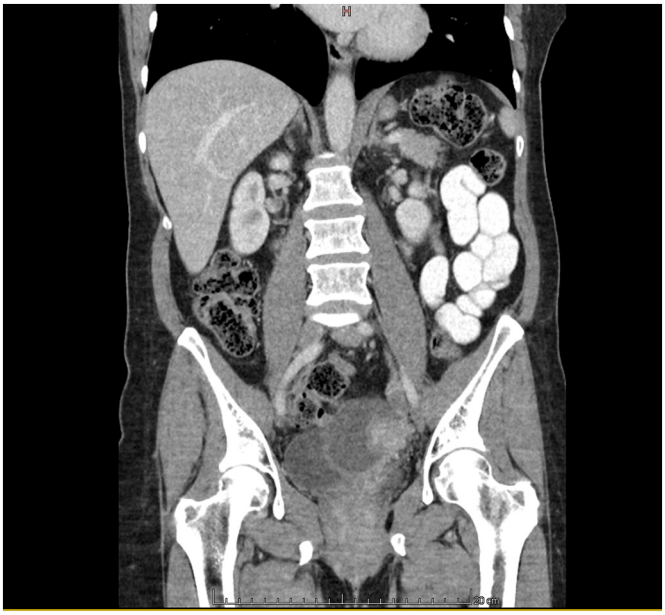

Fig. 1. CT scan showing adnexal mass adjacent to left ovary.

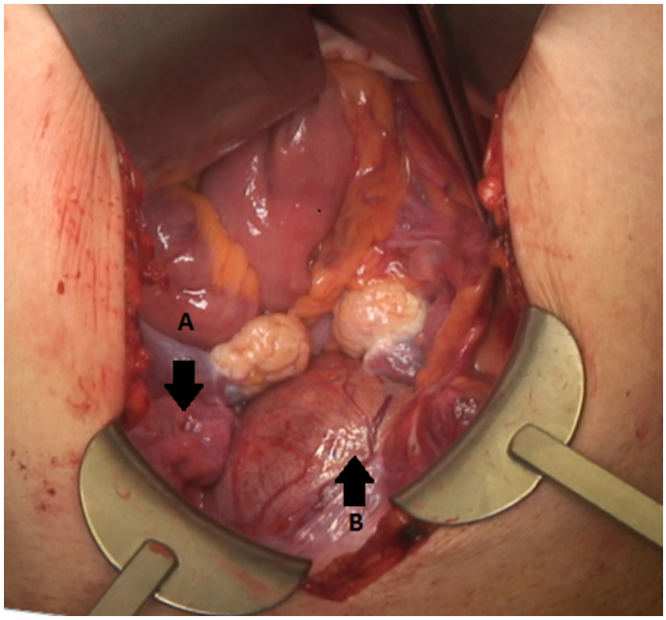

Fig. 3. Intraoperative photograph: A–Uterine deposit B-Residual cervical mass.

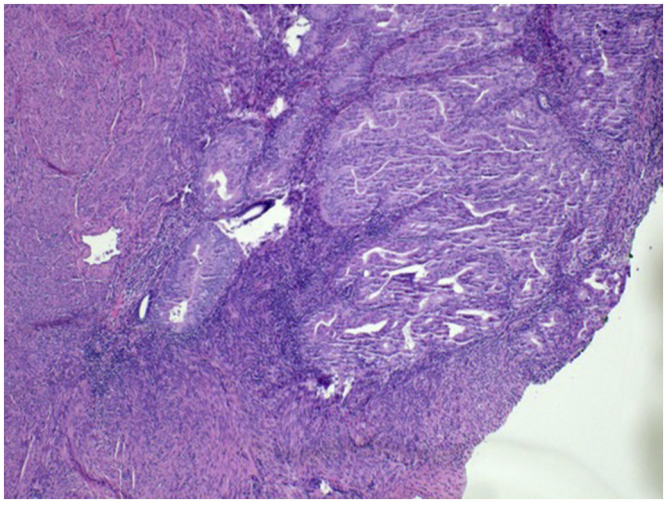

Fig. 4. Histopathology. Grade 1 Endometrioid carcinoma with adjacent myometrial tissue in pelvic mass (hematoxylin and eosin).

developing in uterine deposits following a hysterectomy for benign disease.

Specific risk factors for this patient included the initial subtotal hysterectomy with uterine morcellation and the use of unopposed estrogen hormone replacement. It is likely that the method of initial surgery led to a deposit of uterine tissue implanting within the pelvis and the unopposed estrogen led to promotion of hyperplasia, nuclear atypia, and eventually adenocarcinoma in endometrial tissue within the implant.

While the practice of uterine morcellation has been called into question recently, this case highlights the risk of morcellation, even on benign specimens. The use of containment devices may decrease the potential for seeding of tissue; however, there is no long-term data to substantiate this. Furthermore, this case may indicate a need to reconsider the use of unopposed estrogen as MHT in women with a history of subtotal hysterectomy or uterine morcellation where endometrial tissue may have implanted within the abdominopelvic cavity or persist at the site of transection of the uterine corpus from the cervix. These women should be advised to use continuous combined MHT to reduce the risk of development of endometrial cancer, or, alternatively, non-hormonal therapy for menopausal symptoms, depending on age at menopause [6,7]. Patients should be appropriately counselled on the possible risks and benefits of morcellation for presumed benign disease of the uterus.

Contributors

All authors contributed to the preparation of this case report and reviewed and approved the final manuscript.

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

Obtained.

Provenance and Peer Review

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