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CASE REPORT

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Rare Mullerian adenosarcoma of the uterine cervix arising on a background of endometriosis: A diagnostic challenge with risk of malignant transformation—A case report and review of the current literature

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Key Clinical Message

Endometriosis may contribute to Mullerian adenosarcoma development but makes diagnosis challenging given similar symptoms. Survival benefit has not been definitively shown for chemotherapy, hormonal therapy, or radiotherapy, consolidating surgery as the mainstay treatment. Local excision may be a treatment option for patients with confined tumors wishing to preserve their fertility.

KEYWORDS

adenosarcoma, cervix uteri, endometriosis, fertility, uterus

1 | INTRODUCTION

Mullerian adenosarcomas are mixed tumors characterized by benign epithelial and malignant stromal components.[1–3] They arise most commonly from the uterine corpus but also less commonly from the cervix, ovaries, or fallopian tubes.[4] The most recent WHO classification separates adenosarcoma of the uterus into adenosarcoma of the uterine corpus and adenosarcoma of the uterine cervix, depending on the site of origin.[3]

A National Cancer Database study found that adenosarcoma of the cervix makes up approximately 10% of Mullerian adenosarcomas and accounts for 0.16% of all cervical cancers.[5] Unlike adenosarcoma of the uterine corpus, which usually presents in the post-menopausal period, with a median age of 58 years,[2] adenosarcomas of the cervix are diagnosed more often in younger women, with an average age of 45 years.[5,6] Adenosarcoma of the cervix has also been demonstrated to have a better prognosis and present at an earlier FIGO stage than uterine corpus adenosarcoma.[7]

Here we report the rare case of a 41-year-old woman diagnosed with adenosarcoma of the uterine cervix with a past medical history of endometriosis. We present an overview of adenosarcoma of the uterine cervix and highlight its potential link with endometriosis and the diagnostic uncertainty this may cause. We discuss and acknowledge the challenges in diagnosis and management and the paucity of recommendations around treatment and follow up, including fertility-sparing surgery and the use of radiotherapy, chemotherapy and hormonal therapy. This case adds to the limited literature around presentation and management of this condition, which is especially important when current practice is often based on information or experience with other tumors, such as adenosarcoma of the uterine corpus due to the rarity of this pathology.

2 | CASE HISTORY

We report the case of a 41-year-old nulliparous woman who presented to the tertiary centre endometriosis telemedicine clinic following transfer from a private consultation with heavy and painful periods, and chronic back pain. Review of systems identified no additional symptoms. She had a past medical history of endometriosis and past surgical history of right ankle operation. She was awaiting surgery for endometriosis management. At presentation, she was taking as required per rectal diclofenac for chronic pelvic pain as the first step in the analgesic ladder. She had a body mass index of 28.01 kg/m^2 and had never smoked. She had no relevant family history.

3 | METHODS (INVESTIGATIONS AND TREATMENT)

Her last cervical smear was normal following a previous high-risk human papilloma virus (HRHPV) positive sample and colposcopy and biopsy, which was subsequently normal. All previous cervical smears had been normal. Medical treatments attempted thus far for symptom control included simple analgesia, combined oral contraceptive pill, progesterone only pill, and the Mirena coil. These were all stopped due to the lack of improvement in symptoms as well as the occurrence of unwanted side effects including hair loss, mood disturbance, and menorrhagia.

Pre-operative magnetic resonance imaging (MRI) of the pelvis showed no active or fibrotic endometriosis; however, it revealed an incidental polyp prolapsing into the endocervical canal measuring 15×16×24 mm with hemorrhagic cystic foci (Figure 1A,B). Based on this pre-operative imaging, tumor markers were not requested as there was no suspicion of ovarian, fallopian tube or gastrointestinal malignancy. On admission for surgery, the patient's vital signs were oxygen saturation of 98% on room air, respiratory rate of 18, heart rate of 90, blood pressure of 126/85, and temperature of 36.5°C. The patient underwent hysteroscopy, removal of polyp, Mirena coil removal, and laparoscopic excision of endometriosis. There were multiple pelvic and peritoneal spots of endometriosis seen. The large polyp prolapsing into the endocervical canal was resected, and sigmoid, periureteric and bilateral uterosacral endometriotic nodules were fully excised. The histology of the polyp showed endocervical epithelium on the surface of the polyp and haphazard cystic glands principally lined by benign endometrial epithelium. The glands formed rigid cysts with prominent periglandular stromal cuffing, and there were also phyllodes-like architectural changes, such as glandular compression and polypoidal stromal projections into the lumen (Figure 2). The stroma surrounding the glands showed mild atypia and scattered mitotic figures were identified (Figure 3). There were no heterologous elements, sarcomatous overgrowth or high grade atypia, and no myometrial tissue was identified to assess for invasion. Overall, these results were in keeping with an adenosarcoma of the uterine cervix with low grade features. The histology of other specimens confirmed endometriosis, fibrosis, and a possible functional endometrial polyp. Immunohistochemical staining of the tumor demonstrated that it was 7/8 estrogen receptor positive and 8/8 progesterone receptor positive.

4 | CONCLUSION AND RESULTS (OUTCOME AND FOLLOW-UP)

There were no post-operative complications, and the patient was discharged with oral codeine and paracetamol after an overnight stay in hospital. A multi-disciplinary team (MDT) meeting 3 weeks later discussed the histological findings and planned a computed tomography (CT) of thorax, abdomen and pelvis to look for metastatic disease due to the tumor being low grade and confined to the pelvis. The CT did not reveal any metastatic disease. She was

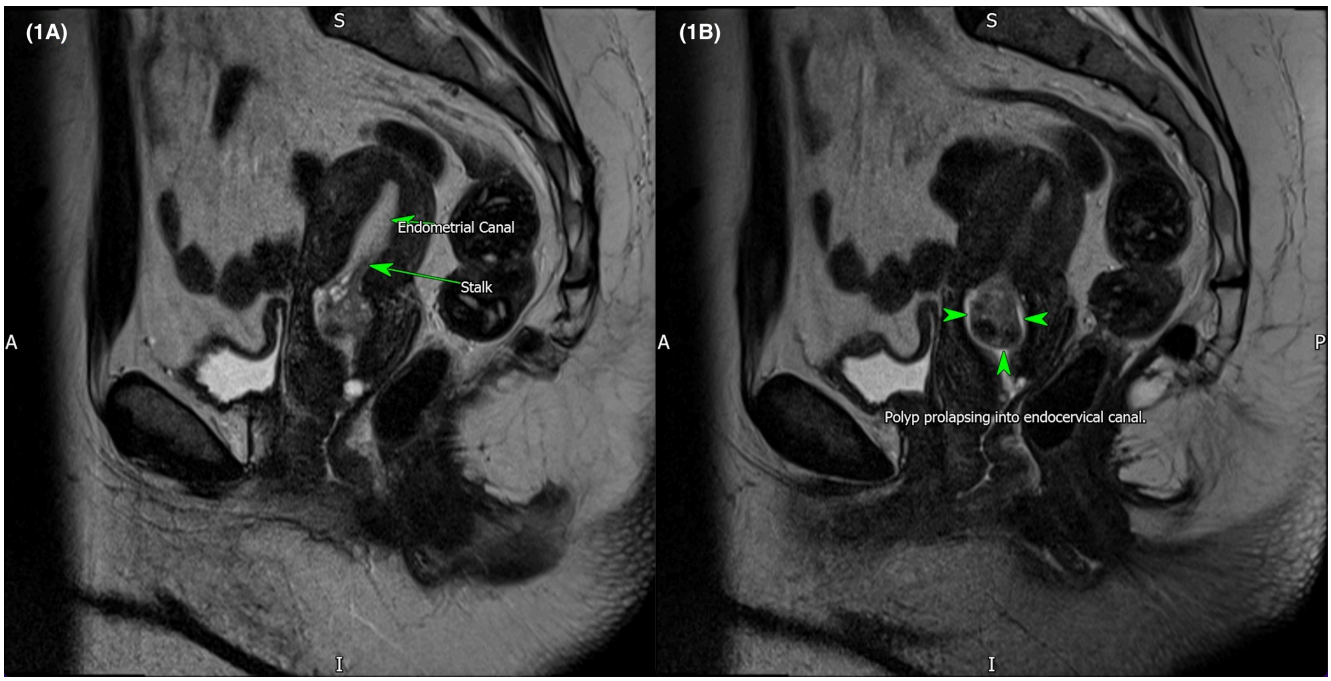

FIGURE 1 Sagittal T2 weighted MRI sequence showing incidental polyp with stalk arising from the lower endometrial cavity (A) and prolapsing into endocervical canal (B).

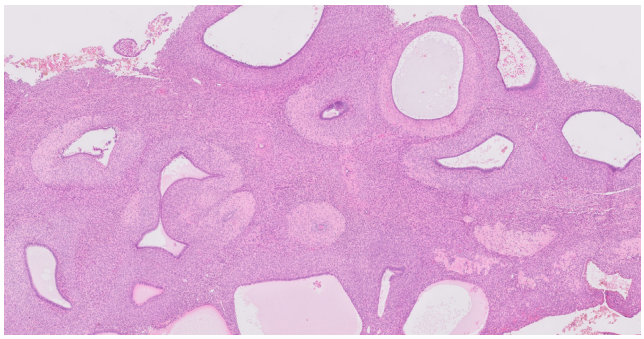

FIGURE 2 Low power view of cervical adenosarcoma showing rigid cysts, phyllodes-like architectural changes and periglandular stromal cuffing.

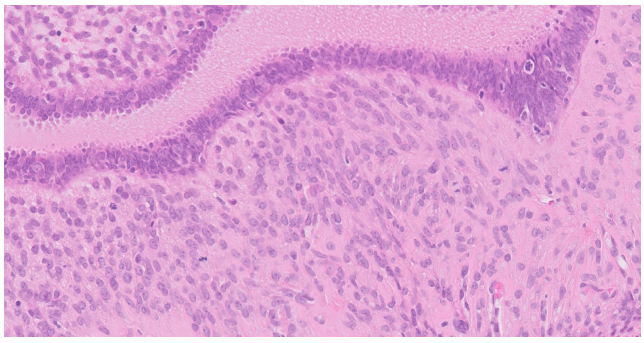

FIGURE 3 High power view of cervical adenosarcoma showing hypercellular stroma with increased mitotic activity and mild atypia.

followed up in the gynecological oncology clinic to discuss completion surgery. The patient was not concerned with fertility preservation as herself and her partner did not want children. A laparoscopic total hysterectomy and bilateral salpingo-oopherectomy was performed under general anesthetic. The evidence of recent surgery was noted, with no malignant disease visible on the peritoneum. However, pelvic and peritoneal endometriosis was seen and confirmed histologically. Histology of the uterus, cervix, ovaries, and fallopian tubes showed no residual adenosarcoma and confirmed FIGO stage 1A adenosarcoma of the uterine cervix based on the previous sample.

On Day 2 post operation, she had a temperature spike of 38.1°C. Blood tests were taken at the time of the temperature spike, which demonstrated a white cell count of 15×10^9/L and normal renal function. Blood cultures had no growth after 5 days. The fever was of unknown cause. Blood tests the following day demonstrated improving inflammatory markers with a white cell count of 11.4×10^9/L. She also had a period of mild hyponatraemia. The patient was discharged on Day 3 post operatively with oral antibiotics.

The patient has since been seen in the gynecological oncology clinic and referred on to see both clinical oncology and menopause specialists. Due to strong tumor expression of both estrogen and progesterone receptors, hormone replacement therapy has been discouraged, with a plan agreed at the MDT to review in clinic regularly with surveillance imaging.

Patient consent was obtained prior to the writing of this case report.

5 | DISCUSSION

Adenosarcoma of the uterine cervix is a rare variant of Mullerian adenosarcoma. Much like endometriosis, this disease presents in pre-menopausal women with symptoms of lower abdominal pain or abnormal vaginal bleeding, including menorrhagia and dysmenorrhoea, as seen in our patient.[2,8–10] This situation can cause diagnostic challenge due to a similarity of symptom profiles of endometriosis and adenosarcoma, and with low-grade tumors mistaken for benign polyps particularly in the younger population, where alternative similar presentations such as endometriosis are more likely.[10] This case demonstrates the importance of ensuring polyps are appropriately investigated even in young patients given their potential for diagnostic uncertainty.

Malignant transformation of endometriosis is known to occur and adenosarcoma secondary to endometriosis has been reported in the literature, most commonly in extra-uterine cases.[11–13] A review of 1000 cases of endometriosis demonstrated that clear cell adenocarcinoma and adenosarcoma are the two neoplasms most associated with extraovarian endometriosis.[14] As well as pelvic malignancies, endometriosis deposits have been associated with gastrointestinal neoplasms of Mullerian adenosarcomatous origin[12,13] in addition to neoplasms of the abdominal wall, umbilicus, and pleura.[15] The mechanism behind malignant transformation of endometriosis is not fully understood, but it has been suggested that oxidative stress from menstruation causes repeated DNA damage.[14] Interestingly, endometriosis may be a positive prognostic factor. Patients with endometriosis associated ovarian cancer usually present earlier, have longer disease-free survival and lower recurrence rates than those with ovarian cancer without endometriosis.[16] However, there is insufficient data to determine whether this applies to other endometriosis associated neoplasms, such as Mullerian adenosarcoma.

The Sampson criteria for malignant transformation of endometriosis dictate that endometriosis sites are found very close to the malignancy, with tumor histology compatible with endometrial origin and that there is no other possible primary tumor observed.[17] The link between adenosarcoma of the uterine cervix and endometriosis presents an interesting hypothesis for the origin of our patient's tumor, which we believe warrants further research. Endometriosis and gynecology oncology teams in Oxford are working to investigate the link between endometriosis and all pelvic cancer, including adenosarcoma. This research involves collecting and analyzing retrospective and prospective data. However, given the rarity of adenosarcoma, prospective studies are challenging. Retrospective studies are likely to provide more information, particularly if they are multicentric.

Regarding cervical adenosarcoma, increased tumor size, increased patient age at diagnosis, myometrial invasion, and sarcomatous overgrowth have been shown to be poor prognostic factors.[5,9] The case discussed shows typical adenosarcoma histology, with the presence of both benign epithelial and low-grade malignant stromal components, both of which are neoplastic, with architecture similar to the leaf-like projections of the phyllodes tumor of the breast.[4] There were no poor prognostic factors identified, particularly no myometrial invasion or sarcomatous overgrowth. The tumor was FIGO staging 1A, indicating that it was limited to the endocervix. Poor prognostic factors associated with adenosarcoma of the uterine corpus in addition to those discussed above for cervical adenosarcoma include cellular atypia, race, resection status, mitosis, and necrosis.[18] These additional prognostic factors may also apply to cervical adenosarcoma, but the small patient population is a challenge to determining this. The presence of a tumor stalk was found to be a protective factor in a retrospective study of both uterine and cervical adenosarcomas.[7]

Case reports of adenosarcoma of the uterine cervix have described a range of tumor grades, from low grade with mild atypia and low mitotic rates[10,19] to high grade sarcomatous overgrowth.[9,20,21] The rarity of adenosarcoma of the uterine cervix makes it difficult to assess the most common presentation and it is a challenge to determine the best treatment options. Currently, the mainstay of treatment is hysterectomy and bilateral salpingo-oophorectomy, rendering the patient infertile. Given that adenosarcoma of the uterine cervix are most common in pre-menopausal women,[22] the option for minimally invasive fertility-preserving treatment in low grade and low stage tumors should be considered but is currently controversial. If fertility preserving treatment is undertaken, robust follow-up including examination and imaging would be necessary to detect recurrence early. Arguments against the use of fertility preserving treatment include the higher risk of recurrence.[23] A literature review by Santiago et al. of 29 cases of low-grade cervical Mullerian adenosarcoma investigated the options for fertility preserving treatment, including trachelectomy and cold knife cone biopsy. Of the 9 patients in this study that underwent fertility preserving treatment, 4 had recurrences, compared to 1 patient out of the 20 whom underwent hysterectomy.[24] This conflicts with another study of 21 cases of cervical adenosarcoma by Yuan et al., which found that fertility preserving treatment was not

associated with worse outcomes in patients with cervical adenosarcoma.[7] None of the 4 patients in this study that underwent fertility preserving treatment experienced recurrence.[7] However, both studies were limited by small patient populations, making it difficult to draw meaningful conclusions.

Data are also lacking regarding the benefit of chemotherapy, hormonal therapy and radiotherapy for Mullerian adenosarcoma. Supporting a role for hormonal treatment of Mullerian adenosarcoma is evidence that hormones, particularly estrogen, could play a role in their development. For example, unopposed estrogen has been linked to the development of gastrointestinal tumors of Mullerian adenosarcomatous origin.[12,13] Several case reports have also described the development of Mullerian adenosarcoma following tamoxifen therapy for breast cancer, although no causal mechanism has been found.[25–29] Tamoxifen is also known to significantly increase the risks of endometrial hyperplasia, polyps, and carcinoma. Interestingly, although it is an antagonist of the estrogen receptor, it has been suggested that it also acts through estrogenic and non-genomic pathways, which could promote cellular proliferation.[30] The action of tamoxifen through estrogenic pathways fueling the development of adenosarcoma would support a role of estrogen in the development of these tumors. Given that breast cancer patients are already at increased risk of endometrial disorders, understanding how tamoxifen increases this risk is important.

Equally, tumor treatment response may be related to estrogen and progesterone receptor positivity.[31,32] Hormonal agents that have been prescribed to patients with uterine adenosarcoma and evaluated in retrospective studies include gonadotropin releasing hormone (GnRH) agonists, such as leuprolide, synthetic progesterones, aromatase inhibitors, and selective estrogen receptor modulators (SERMs). Although these treatments have different mechanisms, they all act to reduce levels of or to block the action of estrogen. Studies have demonstrated that the majority of patients with adenosarcoma of the uterine corpus[7,33] and adenosarcoma of the uterine cervix[18] do not benefit from hormonal therapy, although one retrospective study found that aromatase inhibitor and GnRH agonist therapies improved survival for 2–15 years in 4 out of 28 patients, suggesting a select cohort may benefit.[33] The case discussed here had strong tumor expression of both estrogen and progesterone receptors, which could affect future management decisions in the case of recurrence.

Cytotoxic chemotherapy with doxorubicin-based, platinum-based, trabectedin or gemcitabine regimens has been used to treat recurrent or metastatic Mullerian adenosarcoma, however the evidence for the benefit of these treatments is sparse and largely based on case reports. Additionally, this is mainly related to recurrent or metastatic adenosarcoma of the uterine corpus and not localized disease or adenosarcoma of the uterine cervix. Doxorubicin with ifosfamide has been shown to have superior progression free survival benefit compared to other regimens.[33] Retrospective studies looking at the use of pelvic radiotherapy for Mullerian adenosarcoma have shown no survival benefit[7,18,23] with a study by Seagle et al.[5] showing that adjuvant radiotherapy was associated with decreased overall survival.

In summary, several studies have shown that overall survival is not influenced by adjuvant radiotherapy, chemotherapy or hormonal therapy in adenosarcoma of the uterine corpus[5,7,33] and cervix,[5,7] consolidating surgery as the important mainstay treatment. Adjuvant therapy is therefore generally not recommended but is considered in patients with myometrial invasion, sarcomatous overgrowth and those at high risk of recurrence.

6 | CONCLUSION

In this report, we have documented a rare case of adenosarcoma of the uterine cervix arising on a background of endometriosis.

Early recognition and diagnosis of malignancies associated with endometriosis is essential, particularly in a younger population, when alternative more common diagnoses may be considered in the first instance. This includes investigation of endometrial polyps, which may present diagnostic uncertainty.

A review of the literature surrounding adenosarcoma of the uterine cervix has highlighted a limited evidence base for treatment options, including fertility-sparing surgery, the use of neo-adjuvant or adjuvant chemotherapy and radiotherapy or management with hormonal therapy. Radiotherapy, chemotherapy, and hormonal therapy have not been shown definitively to improve survival in Mullerian adenosarcoma, consolidating surgery as the treatment of choice. Patients with confined tumors who wish to preserve their fertility may be managed with local excision alone provided they are fully counselled about the risk of local recurrence.

This case presents an opportunity to add to the limited literature on management of this pathology, whilst assisting in building a consensus opinion for optimal management, including fertility-sparing protocols, under circumstances where formal guidelines do not exist.

AUTHOR CONTRIBUTIONS

Hannah Bruguier: Project administration; writing – original draft; writing – review and editing. **Natalie**

Maalouf: Writing – original draft; writing – review and editing. **Sarah Smyth:** Supervision; writing – original draft; writing – review and editing. **Stephen Damato:** Data curation; writing – review and editing. **Priyanka Reddy:** Data curation. **Hooman Soleymani Majd:** Conceptualization; supervision; writing – review and editing.

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None.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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