Case report

Cervical Adenosarcoma – Case Presentation with Successful Conservative Management

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Abstract

Adenosarcoma is considered one of the rarest malignancies of female genital system accounting for only about 0.2% of all uterine neoplasms. Due to this rare occurrence, there is a limited amount of data relevant to the treatment and observation of this malignant uterine tumour. Although the treatment in this situation is a total hysterectomy and bilateral salpingo-oophorectomy, it is possible to perform a localized tumour excision if the patient wishes to preserve fertility. It is of paramount importance to monitor the patient due to high recurrence of this tumour despite its most common presentation in a localized stage through abnormal uterine bleeding. In this paper, we present a case of a 34-year-old patient who was admitted because of an abnormal uterine bleeding. Ultrasound evaluation confirmed that there was an endocervical polyp. A localized adenosarcoma was histologically confirmed by endocervical polyp ablation. Regular endometrial and endocervical biopsies have not found any tumor cells and the patient spontaneously conceived and successfully delivered two pregnancies. This case is worth presenting because of its rarity and limited amount of existing data and research on the subject.

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KEYWORDS: uterine sarcoma, adenosarcoma, cervix, gynaecologic neoplasm, abnormal uterine bleeding, polyp

Introduction

Adenosarcoma is a very rare malignancy of female genital system. It represents 5-9% of uterine sarcomas and only 0.2% of all uterine neoplasms (1, 2). Apart from adenosarcoma, there are several different subtypes of uterine sarcomas: leiomyosarcomas, endometrial stromal sarcomas and undifferentiated sarcomas. Carcinosarcomas are also a part of this group and they have been defined as a differentiated form of endometrial carcinoma (2). Unlike carcinosarcomas, adenosarcoma is considered a less aggressive tumour. However, due to its extremely rare occurrence in clinical practice, a limited amount of data and research is available, and consequently, guidelines regarding the monitoring, treatment and prognosis of this tumor. Here we represent a case of a 34-year-old nulliparous patient who wished to preserve her fertility when diagnosed with cervical adenosarcoma. Local excision was performed with close follow-up and the patient conceived successfully two times giving birth to healthy children. Precisely because of its very rare occurrence, this case of a young woman with cervical adenosarcoma is worth presenting.

Case presentation

A 34-year-old patient with no prior gynecological morbidity and a history of regular gynecological check-ups was admitted to the gynecology ward of the county general hospital in early 2014 due to abnormal uterine bleeding. Ultrasound examination revealed a thickened endometrium, and a 10-day course of progestin therapy (dydrogesterone) was prescribed, resulting in a significant reduction in bleeding.

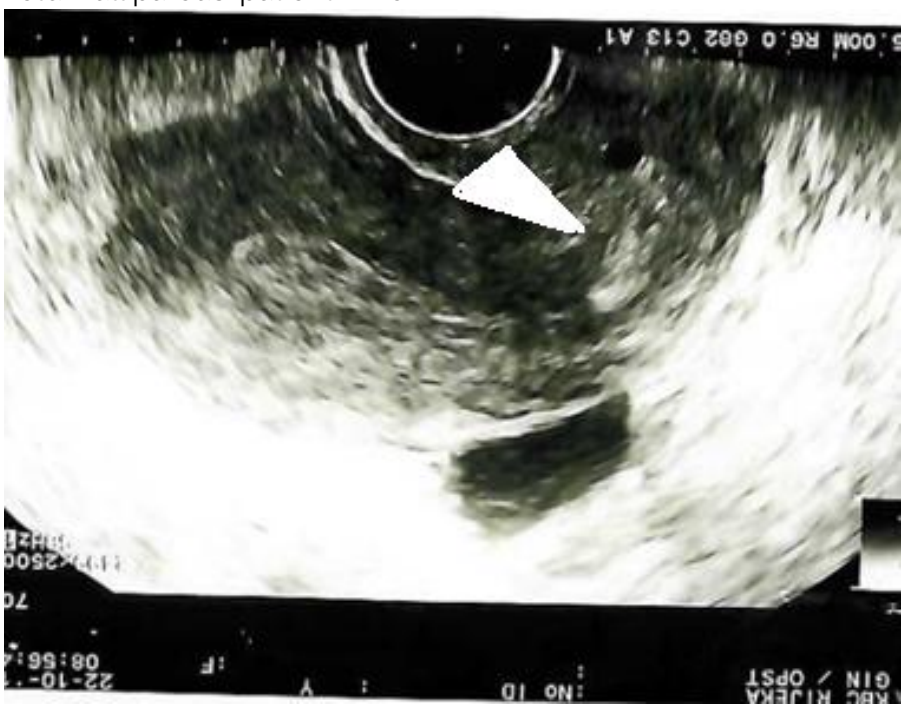

Figure 1. Transvaginal sonography showing endocervical thickening with several anechoic internal cysts (arrowhead) suggesting endocervical polyp

After the bleeding had stopped, the patient was examined. During an ultrasound examination, a dilated cervical canal was noticed (Fig. 1). Also, there was a hyperechogenic formation (3x3x2cm) and increased vascularisation on Doppler which suggested an endocervical polyp. The patient was then admitted to the hospital and several tests and procedures were conducted: Pap smear, endocervical ablation of the polyp, endocervical curettage and endometrial biopsy. Subsequently, the cytodiagnostic result of PAPA was negative for an intraepithelial or invasive lesion, while the pathohistological result sent for consultation to

the Department of Gynecological and Perinatal Pathology of a tertiary care institution indicated an adenosarcoma of the ablated cervical polyp and a normal finding of endocervical curettage and of tissue obtained by endometrial biopsy sample. Furthermore, MRI of the pelvis gave normal results, as did the pulmonary radiogram and ultrasound of the urinary tract. In the same month, she was examined by a specialist oncologist at a tertiary center, and a conservative approach to treatment with regular check-ups was decided, considering the patient's age, nulligravida and desire for a future pregnancy. Two months after the initial symptoms, a whole-body PET/CT was conducted and showed a slightly inhomogeneous uterus around cervix area without any visible signs of enlarged iliac lymph nodes (Fig. 2). Furthermore, pathohistological results of the endocervical curettage and endometrial biopsy performed in a tertiary facility have not shown any cancerous tissue residue. This case was presented at a multidisciplinary oncological consilium where a continued monitoring was suggested via regular diagnostic endometrial biopsies every 6 months. During that period, the patient was eumenorrhoeic and asymptomatic. Every further test presented normal results, including Pap smear.

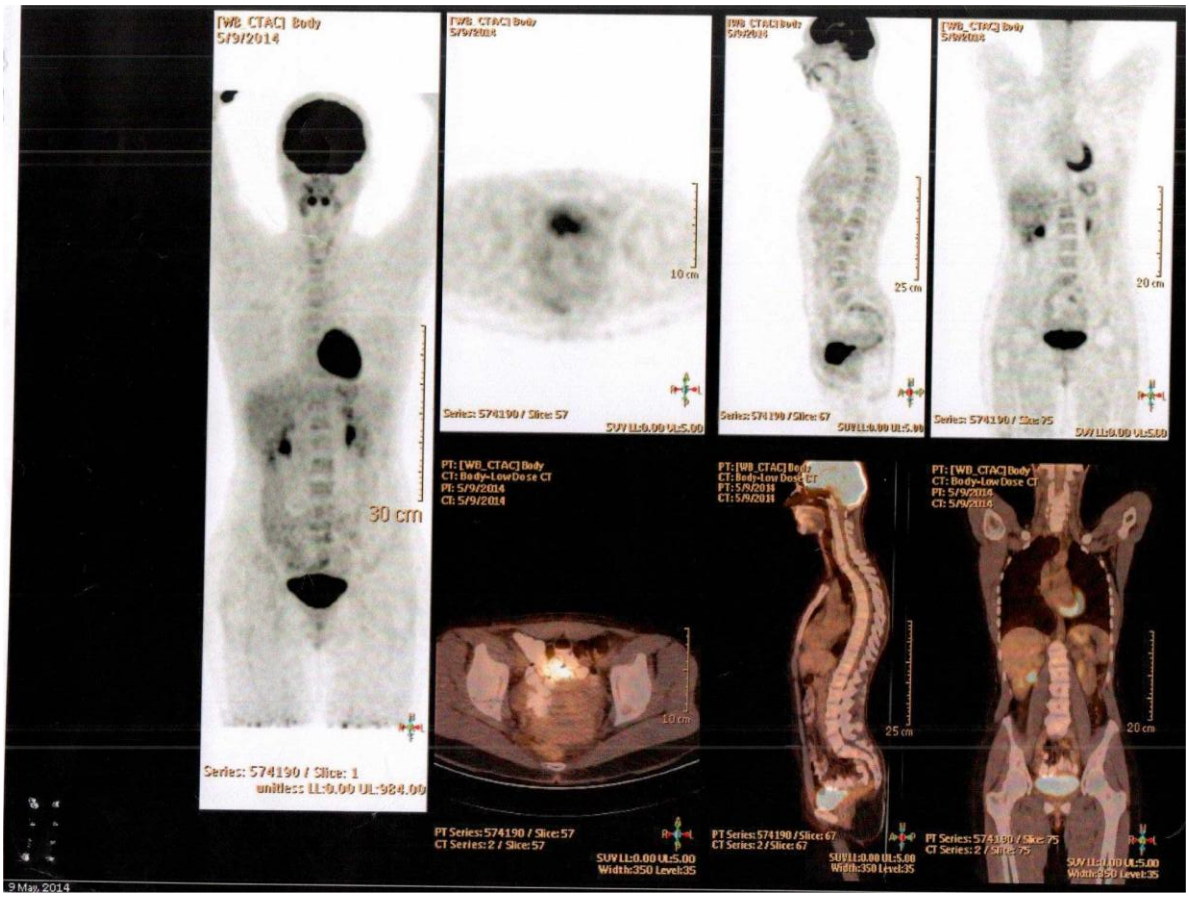

Figure 2. PET/CT scan showing no extent of disease after initial pathological verification of cervical adenosarcoma

One year after the initial symptoms, the patient conceived spontaneously without any difficulties throughout the pregnancy. An induced labour was performed in the 41 week of gestation. However, due to the intrapartum signs of fetal hypoxia, the delivery was finished via caesarean section. The delivered baby was eutrophic and with orderly Apgar score. Three months after the delivery, performed hysteroscopy and endometrial biopsy showed no residue of cancerous tissue. Finally, eight years after the patient was diagnosed with

uterine adenosarcoma, she is in good general health and, with normal pathohistological findings of control biopsies. She conceived spontaneously once again and carried the pregnancy to term giving birth to a healthy child by elective caesarean section.

Discussion

Adenosarcoma represents the rarest form of uterine sarcoma and only about 0.2% of all uterine malignancies(1). According to its patohistological characteristic, adenosarcoma has benign epithelial elements with a malignant mesenchymal component which differentiates adenosarcomas from carcinosarcomas, which have malignant epithelial component (3, 4). The highest incidence of adenosarcomas is later in life, between the ages of 50 and 60, with less than 10% of patients under the age of 40 (1, 5). The majority of these tumours is formed within the uterus and ovaries, followed by cervix, vagina and fallopian tubes, and, very rarely, extragenital areas as peritoneum and omentum (6-8). Risk factors for mostly extragenital adenosarcomas are endometriosis, tamoxifen treatment, previous pelvis radiation and prolonged exposure to estrogen (3, 9-11). Since adenosarcoma usually manifests as a polypoid mass inside the uterine cavity, the most common clinical presentation is an abnormal uterine bleeding which is present in 65-76% of the cases (3, 12). Other symptoms are pain or mass in the pelvis, increased vaginal discharge, discomfort and abdominal distension. They are mostly related to the ovarian adenosarcoma due to its ability to have larger dimensions (13). In the presented case, the ultrasound examination rose suspicion about endocervical polyp after recuperation from an abnormal uterine bleeding and thus directed the patient's treatment towards endocervical excision of the polyp. Although the endocervical polyps are benign, malignant ones can be present in 0.2-1.5% of the cases, mostly in postmenopausal women (14).

Ultrasound evaluation depicts endocervical polyps as well limited and slighly hyperechogenic formations that are flexible when pressure is applied with a probe by Doppler identification of feeding vessels (15). There is a FIGO classification formed in 2009 for uterine adenosarcomas where the presence of myometrial invasion and expansion outside the uterus determines the stage of the disease (16). Most of the patients with adenosarcoma are diagnosed with the disease limited to the uterus – 73.4% (17-20). The treatment for localised adenosarcoma is total hysterectomy (2, 21). However, in patients with no myometrial invasion who wish to preserve their fertility, it is possible to perform a localized excision (22). Given the consideration of tumour spread to adnexa it is recommended to perform bilateral salpingo-oopherectomy (12, 23). The biggest epidemiological study was conducted in Norway and it involved 419 patients with stage I (limited to uterus) of adenosarcoma. It is important to note that approximately one-third of the patients develop local recurrences of the disease over a 10-year period, and due to the aforementioned risk, long-term follow-up of patients is recommended (1, 23, 24). In our case of a younger patient with no medical comorbidities (no endometriosis), the only presenting symptom was an abnormal uterine bleeding and ultrasound examination suggestive of an endocervical polyp. Although there are controversial opinions and guidelines about management of endocervical polyps in premenopausal reproductive age patients concerning regular monitoring or excision, our opinion is that whenever there is an abnormal uterine bleeding and a clinical or ultrasound detected endocervical polyp, the patient should undergo a procedure of polyp ablation and pathohistological analysis should be conducted. Moreover, it is important to visualize the entire uterus during ultrasound evaluation of the genital area and thus pay attention to cervix and endocervical canal.

Conclusion

Considering the rarity of the neoplasm itself, and therefore the available literature, there are still no clear guidelines regarding the monitoring and treatment of this disease. Symptoms such as abnormal uterine bleeding help detect the disease in its early or localized stage.

From this case comes the opinion that every endocervical polyp in reproductive age, whether it is caused by abnormal uterine bleeding or not, should be excised with concomitant endocervical curettage.

Contemporary treatment for this rare malignancy remains total hysterectomy with adnexectomy. However, in the case of limited disease and the patient's desire to preserve fertility, local excision can be performed. Regular follow-up of patients through control biopsies is necessary due to the significantly high recurrence rates described in the available literature.

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Author contribution.

Acquisition of data: DP, MC, VĆ
Administrative, technical, or logistic support: DP, MC, VĆ
Analysis and interpretation of the data: T DP, MC, VĆ
Conception and design: DP, MC, VĆ
Critical revision of the article for important intellectual content: DP, MC, VĆ
Drafting of the article: DP, MC, VĆ
Final approval of the article: DP, MC, VĆ
Guarantor of the study: DP, MC, VĆ
Provision of study material/patients: DP, MC, VĆ

Uloga umjetne inteligencije od prije začeća do postpartalnoga razdoblja

Sažetak

Adenosarkom se smatra jednim od najrjeđih maligniteta ženskog genitalnog sustava, jer čini samo oko 0.2% of svih neoplazmi maternice. S obzirom na takvu rijetku pojavnost, postoji ograničena količina podataka relevantna za liječenje i opservaciju tog malignog tumora maternice. Metoda liječenja u takvoj situaciji je totalna histerektomija uz uklanjanje oba jajovoda i oba jajnika, ali moguće je izvesti lokalno uklanjanje tumora ako pacijentica želi očuvati fertilitet. Od ključne je važnosti nadzor pacijentice uslijed visoke rekurentnosti toga tumora, unatoč njegovu najčešćem prezentiranju u lokaliziranom stadiju kao abnormalno krvarenje maternice. U ovome radu prikazujemo slučaj 34-godišnje pacijentice koja je primljena zbog abnormalnog maternicnog krvarenja. Ultrazvučni pregled potvrdio je postojanje endocervikalnog polipa. Lokalizirani adenosarkom bio je histološki potvrđen ablacijom endocervikalnog polipa. Uobičajene biopsije endometrija i endocerviksa nisu otkrile tumorske stanice i pacijentica je spontano začela i imala dva uspješna poroda. Ovaj je slučaj vrijedan pozornosti zbog njegove rijetkosti i ograničene količine podataka i istraživanja na ovu temu.

Ključne riječi: sarkom maternice, adenosarkom, grlić maternice, ginekološke neoplazme, abnormalno maternično krvarenje, polip