

Uterine Adenosarcoma: A Case Report and Review of Literature

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ABSTRACT

Uterine adenosarcoma (UA) is an infrequent malignancy which contains the epithelium of benign glands and malignant mesenchymal elements. Low-grade adenosarcoma has a sarcomatous part that is uniformly low grade. Uterine adenosarcoma includes 8% of all uterine sarcomas and less than 0.2% of uterine neoplasms. It is more common in perimenopausal or postmenopausal women. Due to the rarity of uterine adenosarcoma, limited information is available to help guide treatment.

A 33-year-old woman, who had one child referred to Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, (Tehran, Iran), with a complaint of abnormal uterine bleeding for one year; the sonography reported a polyp in fundal cavity of uterus. The patient underwent hysteroscopy and curettage. The pathology examination revealed uterine adenosarcoma which led to a total hysterectomy and bilateral salpingo-oophorectomy.

Abnormal uterine bleeding is the most common symptom of UA. At a young age there is a possibility of misdiagnosis. Hysteroscopy should also be performed if symptomatic or enlarge polypoid lesions are diagnosed preoperatively.

Keywords: Abnormal uterine bleeding, Polyp, Uterine adenosarcoma



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Introduction

Adenosarcoma is an infrequent tumor which is recognized as a biphasic tumor composed of both sarcomatous stroma and benign epithelium. Uterine adenosarcoma (UA) accounts for 5.5–9.0% of uterine sarcomas and about 1% of female genital tract neoplasms (1). Due to the rarity of UA, limited information is available to help the physicians, and the available data are based on retrospective small case series and population-based analysis. Here is a case of uterine corpus adenosarcoma in a woman of childbearing age.

Case Presentation

A 33-year-old woman gravida 1, para 1 presented with abnormal uterine bleeding for one year before referring. The cytological test of the uterine cervix was negative. Vaginal examination revealed an enlarged uterus and an ultrasound scan indicated a 30×20 mm polyp in the uterine cavity.

There was a 32×1.5 cm polypoid mass in the fundal cavity of the uterine in hysteroscopy, then resection of the mass was performed and adenosarcoma was reported by two experienced gynecologic pathologists. Final pathological evaluation revealed a low grade mixed mesenchymal tumor composed of a low-grade sarcomatous component (Figures 1, 2, 3). Immunohistochemical study was positive for CD10, SMA, Desmin, and h-Caldesmon. These tumoral tissues were covered by epithelial cells which were positive for pancytokeratin (PCK).

An abdominopelvic CT scan was normal. The chest CT scan didn't show any significant pathological findings.

Total abdominal hysterectomy with bilateral Salpingo-oophorectomy and peritoneal washing was performed. At laparotomy, the uterus was about 8 weeks without any gross residual tumoral mass in the uterine specimen. Both ovaries were intact. Lymphadenectomy was omitted. The histopathological final report

didn't show any residue and revealed secretory endometrium and endometrial polyp with chronic cystic and polypoid cervicitis. Peritoneal washing was negative for malignancy. Her postoperative period was without problem and she was discharged without postoperative treatment and no recurrence was reported in an 8 years follow-up.

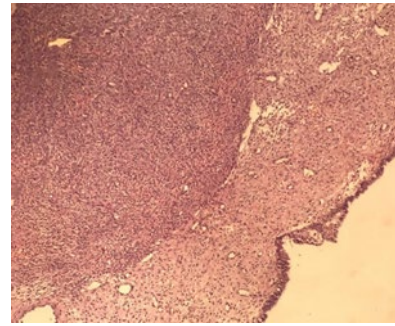


Figure 1. Photomicrograph of uterine adenocarcinoma showing benign looking glandular type epithelium in lower right and neoplastic stromal component in the upper left (H&E staining×40)

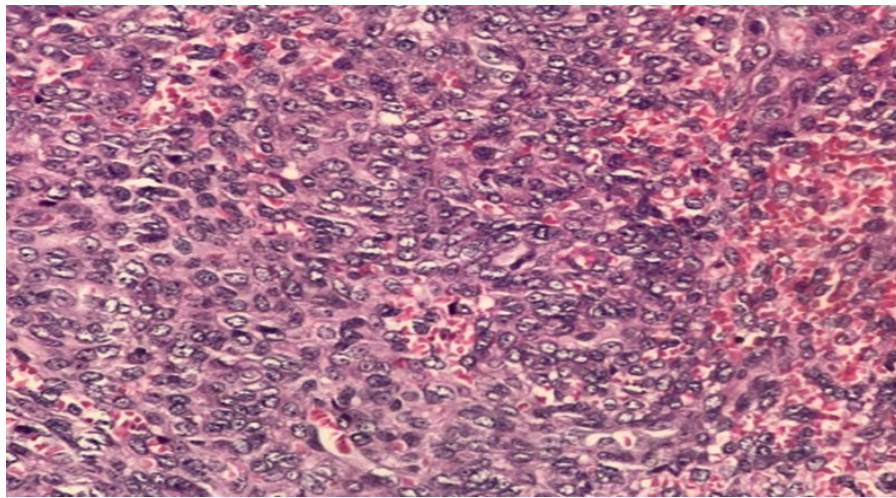


Figure 2. Photomicrograph from uterine adenocarcinoma showing cellular neoplastic stroma with nuclear atypia (H&E staining×100)

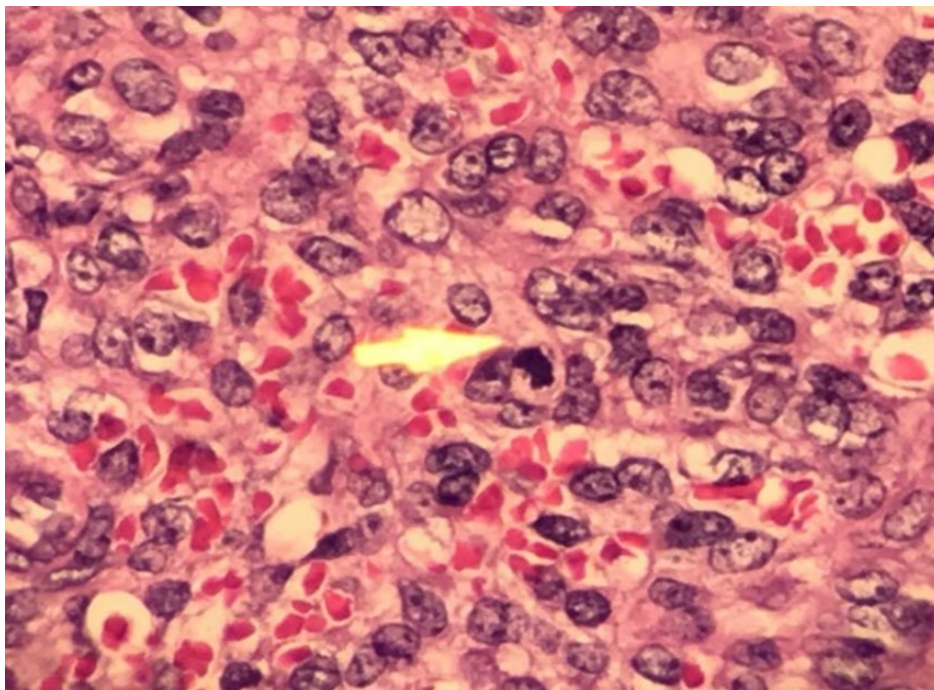


Figure 3. Photomicrograph: stromal component of UA showing mitotic figure (arrow head), marked cellular atypical and pleomorphism are noted (H&E staining×400)

Discussion

Müllerian adenosarcoma was first reported and explained by Clement and Scully in 1974 as a mixed uterine tumor (2, 3). Three types of adenosarcoma have been explained by Soslow and Longacre. Low-grade adenosarcoma has a sarcomatous part that is uniformly low grade. Morphologically, high-grade adenosarcoma has a high-grade sarcomatous section, and sarcomatous overgrowth accounts for 25% or more of the tumor, which is defined as pure sarcoma, containing about 10% of cases (3). The epithelial components are usually similar to endometrial cells but may also resemble secretory, squamous, clear, or mucinous cells (4). Adenocarcinoma may rarely be seen in the epithelial part of an adenosarcoma. In about 25% of cases, heterologous mesenchymal components may be found. Müllerian adenosarcomas are not histologically graded, according to the present World Health Organization (WHO) definition. Adenosarcoma may be considered low in terms of malignant potential and is comparable to the low-grade endometrial stromal sarcoma (5). Uterine adenosarcoma includes 8% of all uterine sarcomas and less than 0.2% of uterine neoplasms (6). It is more common in perimenopausal or postmenopausal women, but some cases have been found in childhood and the age range is reported to be between 13 and 94 (7, 8). In our case, she was 33 years old.

Risk factors for uterine adenosarcoma include endometriosis, adenomyosis, previous pelvic radiation and tamoxifen use, fatness, and diabetes, long-term oral contraceptive use (4), but our patient did not have any predisposing factors.

Some researchers have classified endometrial polyps larger than 1 cm as "large polyps" and those larger than 4 cm as "giant polyps" which are very rare (9). Polyps larger than 1 cm are associated with malignancy, which was concluded by Wong *et al.* (10). The number of polyps and the volume of polyps more than 10 cc is associated with neoplasm of endometrial polyps, which Elfayomy and Suleiman mentioned (11).

Irregular uterine bleeding is the most common symptom of adenosarcoma and is comparable to other types of uterine sarcomas. If adenosarcoma occurs in premenopausal women, it usually presents with irregular menstrual cycles and sometimes enlargement of the uterus (e.g. current literature), and symptoms of pelvic mass are seen.

TAH without BSO in premenopausal women is an option. Lymphadenectomy is not essential in patients with the limited uterine disease without high-risk factors (12). There is no optimal adjuvant systemic treatment strategy, but standard sarcoma chemotherapy regimens may be effective in adenosarcoma and adeno-

sarcoma with sarcomatous overgrowth (13). Chemotherapy may be given to patients with the recurrent or unresectable disease (14).

Most patients with adenosarcoma present with stage I disease and their overall 5-year survival is 60-80%. This case also was presented in stage I. The mortality rate of adenosarcoma without sarcomatous overgrowth has been revealed to be between 10% - 25%, whereas for adenosarcoma with sarcomatous overgrowth it may be up to 75% (3).

Poor prognostic factors include sarcomatous overgrowth, extensive myometrial invasion, presence of heterologous parts, and extrauterine involvement (15).

The recurrence rate of adenosarcoma without sarcomatous overgrowth is between 15% - 25%, but in patients with sarcomatous overgrowth up to 45-70% has been reported. Extensive myometrial invasion, lymph node involvement, heterologous stromal parts, and beyond uterine manifestation are associated with increased recurrence rates.

Recurrence occurs in approximately 50% of patients which is usually seen in the vagina and pelvis. In about 5% of the cases, distant metastases have been described, for example in lungs (3). Fortunately, in this case, after 8 years of follow-up, she has remained without any signs and symptoms of recurrence.

Conclusion

Abnormal uterine bleeding is the most common symptom of UA. At a young age there is a possibility of misdiagnosis. Abnormal uterine bleeding in patients might be the result of cancer. Hysteroscopy should also be performed if symptomatic or enlarge polypoid lesions are diagnosed preoperatively.

Due to the rarity and non-specificity of clinical and paraclinical manifestations, they often lead to misguidance. Therefore, identifying the risk factors associated with this disease and conducting effective follow-up and intensive screening for high-risk patients as well as related interventions for high-risk factors is critical.

Acknowledgments

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Conflict of Interest

The authors declared no conflict of interest.

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