Uterine adenosarcoma diagnosed following hysteroscopic resection of an intrauterine tumour

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Summary

Uterine adenosarcoma is a mixed müllerian tumour consisting of a benign epithelial component and a malignant stromal component. It is a rare tumour that represents 8% of uterine sarcomas. We present a case of a 61-year-old woman who underwent a surgical hysteroscopy for postmenopausal metrorrhagia and thickened endometrium detected by ultrasonography. The pathologic diagnosis of the tumour removed by hysteroscopy was uterine adenosarcoma. The description of this case provides an opportunity to review the literature on uterine sarcomas diagnosed by surgical hysteroscopy.

Key words: Uterine sarcoma; Surgical hysteroscopy; Adenosarcoma.

Introduction

Uterine sarcomas are rare, representing 3% of malignant tumours of the uterus [1]. Clinical diagnosis of sarcoma is difficult. Generally these tumours present as growths indistinguishable from myomas and occur in 0.13 to 0.30% of leiomyosarcomas [2, 3] or as an intracavitary growth suggestive of a polyp or submucosal myoma, which may occasionally exit the uterus via the cervical orifice [1]. The malignant transformation of an endometrial polyp or a myoma occurs in fewer than 5% and 1% of cases respectively [4, 5].

Hysteroscopy makes it possible to treat uterine polyps and myomas conservatively [6, 7] with minimally invasive outpatient surgery, and to diagnose hysteroscopically a malignant uterine tumour.

A case of uterine adenosarcoma diagnosed by surgical hysteroscopy is presented together with a review of the cases described in the literature.

Case Report

A 61-year-old woman presented at our hospital for postmenopausal metrorrhagia over a period of two weeks, with bleeding in quantities similar to her menstrual periods.

Her family history included stomach cancer and breast cancer. Her medical history included diabetes mellitus managed with dietary therapy, slight hypertension and hepatitis B. Her gynaecological and obstetric history revealed menarche at age 12, regular menstrual cycles, three normal deliveries, and menopause at age 52. In December 1994 she presented with postmenopausal metrorrhagia over a period of ten days. Upon examination an endocervical polyp and thickened endometrium (10 mm) were detected by ultrasonography. Hysteroscopy revealed polypoid formations which were removed with scissors along with the endocervical polyp. The pathologist's report confirmed the presence of an endocervical polyp measuring 7 x 6 mm and an endometrial polyp measuring 13 x 5 mm.

In August 2003 this woman presented again with metrorrhagia. Ultrasonography showed a polypoid mass protruding through the cervix, and an endometrial thickness of 22 mm. A second hysteroscopy was performed revealing an endometrial polyp occupying the entire uterine cavity and a cervical tumour, both of which were removed with scissors. The pathologist's report confirmed the presence of an endocervical polyp measuring 25 x 10 mm, and an endometrial polyp measuring 45 x 18 mm, both of which were benign.

In June 2004 the results of the gynecological examination were negative, and vaginal ultrasonography showed an enlarged uterus measuring 94 x 54 x 71 mm and an endometrial thickness of 40 mm, heterogeneous and irregular with vascularization present (Figure 1). A hysteroscopy was scheduled. It revealed a mass occupying the entire uterine cavity with a broad attachment to the posterior wall which was removed almost entirely by hysteroscopic resection, fragmenting it with a cutting loop and using 1.5% glycine solution for uterine distention. The surface of the mass was covered with endometrial tissue but when sliced with the cutting loop its consistency was hard and suggestive of a uterine myoma. The procedure was performed on an outpatient basis under paracervical block and sedation.

Macroscopic analysis carried out by the pathology laboratory showed histoid fragments of elastic consistency weighing a total of 26 g. The histopathology indicated a mass consisting of both epithelial and stromal components. The epithelial component was benign and endometrial and the stromal component consisted of proliferation of fusiform cells which, in some areas and surrounding certain glandular structures, showed a greater cellular density with slightly or moderately cytological atypia the presence of mitotic figures in numbers from 2 to 4 per 10 high-power fields (Figures 2, 3 and 4). The immunocytochemical tests showed desmin focally positive in the stromal component and elevated Ki67 in the hypercellular areas.

The final diagnosis was uterine adenosarcoma, and in consequence the patient underwent hysterectomy with bilateral salpingo-oophorectomy and peritoneal lavage. Peritoneal cytology was negative and analysis of the surgical specimen showed a uterine cavity that was distended and occupied by a polypoid mass with a broad area of attachment measuring 30 x 20 mm.

The final diagnosis of the surgical specimen was adenosarcoma originating in an endometrial polyp and invading its attachment to the uterine wall. The tumour did not invade any of the other structures analyzed.

The patient's postoperative course was normal and the hospital's tumour board decided not to use adjuvant therapy.

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Fig. 1

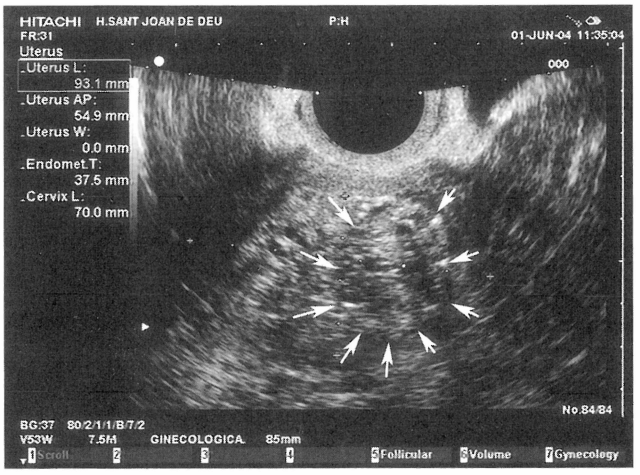

Fig. 2

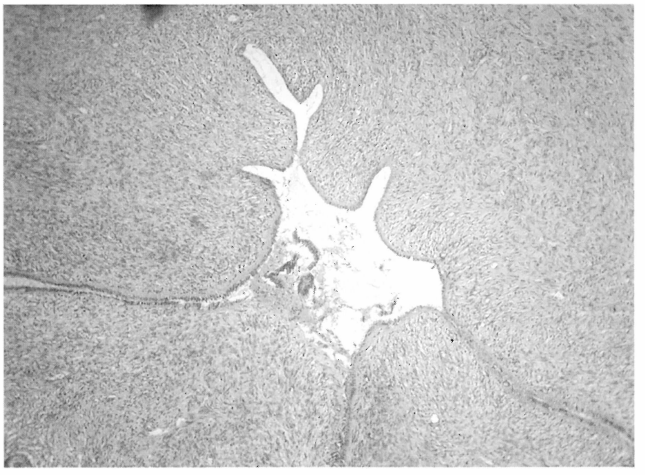

Fig. 3

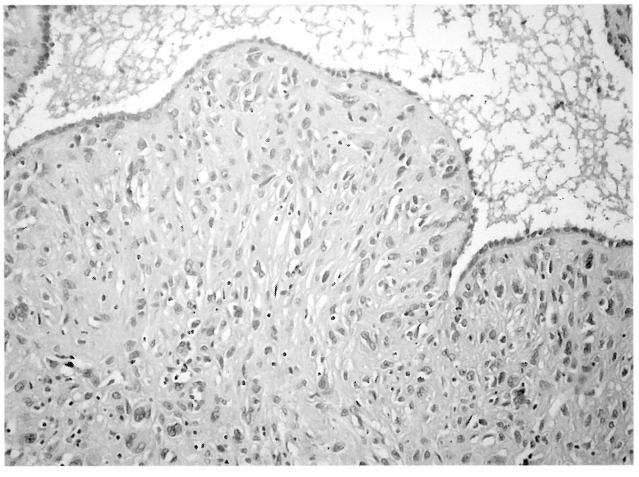

Fig. 4

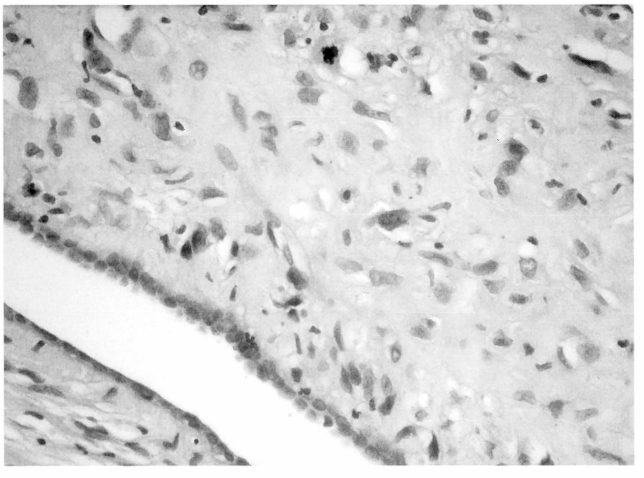

Figure 1. — Intravaginal ultrasound with arrows indicating endometrial thickness of 40 mm.
Figure 2. — Glandular structure of foliaceous appearance as a consequence of stromal hypercellularity.
Figure 3. — Glandular structure covered with benign epithelial cells and showing atypical hypercellular stroma.
Figure 4. — Presence of mitosis in the stroma.

Discussion

Uterine adenosarcoma is a mixed müllerian tumour characterized by its composition, consisting of a benign epithelial component and a malignant stromal component. It was first described by Scully [8], and it is a rare tumour representing 8% of uterine sarcomas [9]. It originates in the endometrial cavity but it may also appear in the cervix as well as in the vagina, the myometrium, ovaries, or pelvic cavity in areas where endometriosis develops [10].

Uterine adenosarcomas may appear at any age (14-89 years) [1, 11], but tend to do so more often in older women. The average age at presentation in extensive series is 58 years, similar to our case (61 years) [11]. The initial symptom with which these tumours present is metrorrhagia [11, 12], and they tend to appear as exophytic polypoid masses growing inside the uterine cavity, as in our case. Microscopically they consist of a benign epithelial component of endometrial cells and a sarcomatous component that tends to be hypercellular in periglandular areas, resulting in a foliaceous appearance. It is in these areas that cytological atypia is greatest, and the greatest number of mitotic figures is observed (> 2 per 10 high-power fields). The stromal component may consist of homologous elements derived from the uterus itself. Alternatively, and less frequently, it may be heterologous, and the sarcomatous elements are not part of the uterus.

Ultrasonography plays a fundamental role in diagnosing endometrial pathology, as demonstrated by our case, in which the thickness of the endometrium (40 mm) was clearly pathological in a postmenopausal woman [13, 14].

In this case, given the patient's history of benign polyps and her desire to preserve her uterus, a hysteroscopy was scheduled because a new polyp was suspected, although its rapid growth was a cause of concern; only a year earlier, a benign endometrial polyp had been removed.

A review of the literature shows that a not inconsiderable number of cases of malignant uterine tumours are diagnosed following surgical hysteroscopy [15-19].

If we focus exclusively on sarcomas, leaving aside endometrial carcinomas which are the most frequent gynaecological cancers in developed countries, the number of cases decreases significantly.

The sarcomas described in the literature are diagnosed following endometrial ablation [20-22] or removal of a

mass whose appearance in ultrasound results is suggestive of submucosal myoma [16, 23]. In extensive series of 800 and 2,402 endometrial ablations, one and three uterine sarcomas were detected, respectively [20, 22]. Also described in the literature are cases of resection of supposed myomas by hysteroscopy in which the histologic diagnosis was sarcoma [18, 23, 24]. This is similar to our case in which the hysteroscopic image of the mass was suggestive of submucosal myoma or a polyp with a broad base of attachment.

With respect to histologic type, the literature indicates that endometrial ablation and myoma resection are rarely associated with a diagnosis of sarcoma of the endometrial stroma, leiomyosarcoma, and carcinosarcoma, although it is impossible to say whether any of these histologic types are more frequent because of the small number of cases [16-24]. We have not found any case of adenosarcoma diagnosed by surgical hysteroscopy in the literature.

We did find reports of hysteroscopic diagnosis of malignant mixed müllerian tumours in the literature [25], although this type of diagnosis is probably easier since both the epithelial and stromal components are malignant.

The risk of malignancy increases with the age of the patient, although in the literature there are reports of cases in premenopausal women [18, 21, 23].

Hysteroscopy is not counterindicated in the case of malignant tumours unless a diagnosis of malignancy has already been made. Surgical hysteroscopy makes accurate histologic diagnosis possible, which in turn facilitates appropriate surgical intervention. In the absence of a previous diagnosis, a second surgery may be required in order to complete treatment.

The standard treatment for uterine sarcomas is total hysterectomy with bilateral salpingo-oophorectomy and peritoneal lavage [1, 24], as in the present case. In these tumours the prognosis depends on the number of mytotic figures present and the degree of cytological atypia, sarcomatous overgrowth in more than 25% of the tumour; deep invasion of the myometrium; the presence of heterologous components; and the results of peritoneal cytology [1, 26]. In our case the prognosis was good and no adjuvant therapy was used since the peritoneal cytology was negative and the tumour presented few mitotic figures (2 to 4 per 10 high-power fields), slight or moderate cytological atypia, no deep invasion of the myometrium, only one area of stromal overgrowth and no heterologous component.

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