CASE REPORT

Endometrial adenocarcinoma in one horn of a didelphic uterus with vaginal duplication

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SUMMARY

A 59-year-old female patient presented with vaginal bleeding. A didelphic uterus with vaginal duplication was diagnosed on the basis of physical examination and radiology tests. Biopsy revealed an endometrial cancer in the left horn, while the right was atrophic. Laparoscopic hysterectomy, bilateral salphingo-oophorectomy, pelvic and para-aortic lymphadenectomy were performed. According to Federation International of Gynecology and Obstetrics (FIGO) staging the tumour was classified Ib. The adjuvant therapy was vaginal cuff brachytherapy. After 6 months, she has no evidence of the disease.

BACKGROUND

Cancer of the corpus uterine is the sixth most common malignancy in women worldwide with an estimated incidence of about 287 100 new cases in the year 2008, solely.¹

In developed countries, endometrial cancer is the most common malignancy of the female genital tract and the fourth most common cancer in women. In the USA there were 46 470 new cases and 8120 deaths from the disease in 2011.²

Gynaecologists should keep in mind the Müllerian duct anomalies in the differential diagnosis of postmenopausal bleeding, because if not, an endometrial cancer could be overlooked.

There are few cases published in the literature about endometrial cancer in patients with uterine malformations. To the best of our knowledge, this is the first case of vaginal duplication.

CASE PRESENTATION

A 59-year-old female patient presented to her primary care physician with postmenopausal bleeding. Her obstetric history consisted of two caesarean sections.

She was referred to our hospital. Physical examination revealed two different vaginas and two cervixes.

INVESTIGATIONS

Transvaginal sonography revealed a didelphic uterus. The thickness of the endometrial images was 16 and 4 mm. Hysteroscopy showed images that were suspicious of malignancy (excrecent tumour) in the left horn, and atrophic images of the right one. The result of the biopsy was an endometrioid adenocarcinoma grade 1. MRI confirmed a didelphic uterus and showed that myometrial infiltration was more than 50% (figure 1). CT

Figure 1 MRI of the pelvis with didelphic uterus.

showed that the disease was confined to the uterus. There were no enlarged lymph nodes.

DIFFERENTIAL DIAGNOSIS

Postmenopausal bleeding can be caused by: vaginal atrophy, endometrial hyperplasia, endometrial polyps, cervical polyps and endometrial carcinoma. Although the most frequent cause is vaginal atrophy, it is always mandatory to exclude an endometrial carcinoma.

TREATMENT

The sole relevant finding in the operation room was the didelphic uterus (figure 2).

The surgical techniques used were laparoscopic hysterectomy, bilateral salphingo-oophorectomy, pelvic and para-aortic lymphadenectomy. Six trocars were used: (1) the Hasson trocar which was placed just below the umbilicus, (2) a 10 mm supra-pubic trocar and (3) four trocars of 5 mm, two on each side. In this kind of hysterectomy, performed in a malformed uterus, the ureters could be much closer to the uterus than usual. In order to avoid

Figure 2 Laparoscopic view showing didelphic uterus.

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Figure 3 Laparoscopic view showing vaginal duplication.

Figure 4 Hysterectomy specimen.

ureteral injury both ureters were dissected from the iliac vessels to the bladder. As she had had two caesarean sections, the dissection of the bladder was made very carefully. The uterine manipulator used was Rumi which was placed in the left vagina (figure 3). To identify the right vagina a gauze pad was entered. Both vaginas were opened with monopolar energy. She was discharged after 2 days.

Gross examination revealed a didelphic uterus (figure 4). The left horn was 7.8×4.5×3 cm. The right horn was 5.5×2.8×2.3 cm. Microscopical examination revealed that the tumour occupied the entire endometrial cavity of the left one, and myometrial infiltration was more than 50%. There was no lymphovascular invasion. The final histological examination confirmed an endometrioid adenocarcinoma in the left horn, tumour grade 1. The endometrium of the right uterine horn was atrophic. The pelvic lymph nodes were free of tumour (0/34), also the para-aortic nodes were free of tumour (0/26). The tumour was staged Ib Federation of Gynecology and Obstetrics (FIGO).

OUTCOME AND FOLLOW-UP

According to the PORTEC³ criteria the adjuvant therapy was vaginal cuff brachytherapy.

The surgery was performed 6 months ago, and at the time of writing this case report the patient was without evidence of the disease.

DISCUSSION

When a postmenopausal patient has vaginal bleeding it is always necessary to exclude an endometrial carcinoma. If the patient has a didelphic uterus, both horns have to be examined. Performing a biopsy on just one of them could overlook the tumour, delaying the diagnosis and treatment of the patient.

The Müllerian anomalies have not been implicated as a significant risk factor for the development of an endometrial cancer.

Table 1 Review of literature

Ref	First author	Year	Age of patient	Uterine malformation	Tumour type and grade	Primary treatment	Adjuvant treatment	Outcome
4	Kondi-Pafiti A	2002	58	Didelphic	Endometrioid adenocarcinoma, G3	Abdominal hysterectomy with bilateral salpingo-oophorectomy	Chemotherapy and radiotherapy	Well after 1 year
			49	Bicornuate	Adenocarcinoma, G1	Abdominal hysterectomy with bilateral salpingo-oophorectomy	No treatment	Well after 2 years
			76	Bicornuate	Endometrioid adenocarcinoma, G1	Abdominal hysterectomy with bilateral salpingo-oophorectomy	No treatment	Well after 6 months
5	Moore JR	2004	74	Bicornuate	Mixed endometrioid and serous papillary carcinoma, G2	Abdominal hysterectomy with bilateral salpingo-oophorectomy, omentectomy, pelvic and para-aortic dissection	Chemotherapy and vaginal cuff brachytherapy	No information available
6	Molpus KL	2004	66	Didelphic	Adenocarcinoma with clear cell features, G3	Abdominal hysterectomy with bilateral salpingo-oophorectomy	Chemotherapy and radiotherapy	No information available
			59	Didelphic	Adenocarcinoma, G1	Abdominal hysterectomy with bilateral salpingo-oophorectomy and excision of the vaginal septum	No treatment	No information available
7	Fanfani F	2006	33	Didelphic	Endometrioid adenocarcinoma, G1	Abdominal hysterectomy with bilateral salpingo-oophorectomy and omentectomy	No treatment	No information available
8	Iavazzo C	2013	73	Didelphic	Uterine Carcinosarcoma, G3	Abdominal hysterectomy with bilateral salpingo-oophorectomy	Radiotherapy	No information available

While infertility is clearly associated with uterine malformations, the association with neoplasia is extremely rare. Moreover, there are very few cases published in the literature^{4–9} (table 1). As far as we know, this is the first case report of a patient with a didelphic uterus and vaginal duplication.

The incidence of congenital anomalies of the uterus is about 4–5%, although it may probably be higher, because many are clinically silent.¹⁰ There are different types of anomalies: septate (35% of the cases), bicornuate (26%), arcuate (18%), unicorniate (10%), didelphic (8%) and agenesis (3%). Uterine didelphis happens when there is an incomplete fusion of the Müllerian ducts. Duplication is limited to the uterus and cervix, although duplication of the vulva, bladder, urethra, vagina and anus may also occur.¹¹

The standard surgery of an endometrial cancer is radical type A hysterectomy (total extrafascial hysterectomy)¹² with bilateral salpingo-oophorectomy. The pelvic and para-aortic lymph node dissection is carried out if there are risk factors.

In our hospital, according to the international criteria, the indications of lymphadenectomy in endometrial cancer are: grade 3, non-endometrioid, myometrial invasion >50%, tumour >2 cm.

Learning points

- ▶ Endometrial cancer must always be excluded when a postmenopausal patient has vaginal bleeding.
- ▶ If a patient with a didelphic uterus has postmenopausal bleeding, both horns must be examined.
- ▶ When a gynaecologist is performing hysterectomy of a malformed uterus, the ureters should be dissected in order to avoid ureteral injury.

In our point of view, this surgery has to be performed by a gynaecological oncologist with experience in laparoscopic surgery as ureters could be much closer to the malformed uterus.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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