

A single horn endometrial carcinoma of a uterus bicornis unicollis

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In this report, we describe a case of endometrial carcinoma arising in one horn of a bicornuate uterus. The diagnosis of this rare combination can be missed unless an unrecognized postmenopausal bleeding alerts the gynecologist to make a careful search for both endometrial cavities that may be curetted. Physicians should remember the possible existence of a separate uterine cavity when endometrial cancer is clinically suspected but histology fails to confirm the diagnosis.

Key Words: Endometrial cancer, Bicornuate uterus, Single horn carcinoma, Postmenopausal bleeding

INTRODUCTION

Most uterine anomalies result from a defect in the development or fusion of the paired müllerian ducts during embryogenesis. The incidence of these abnormalities is 0.16% to 10% in the general population, but this number might be underestimated.¹ In fact, some patients can be asymptomatic, and the diagnosis is often postponed after a gynecologic examination for sterility, infertility, or repetitive spontaneous abortions.

A bicornuate uterus results from failure of the müllerian ducts to completely fuse. The central myometrium may extend to the level of the internal cervical os (bicornuate unicollis) or external cervical os (bicornuate bicollis). The latter is distinguished from uterus didelphys because it demonstrates some degree of fusion between the two horns, whereas in classic uterus didelphys the two horns and cervixes are separated completely.²

There are only a few cases reported of an endometrial carcinoma arising in patients with müllerian ducts abnormalities, and no correlation has been ever shown between the presence of an uterine malformation and endometrial cancer risk.³⁻⁵ In this report, we describe a case of endometrial carcinoma arising in one horn of a bicornuate uterus.

CASE REPORT

A 58-year-old obese woman, gravida 3, para 3 presented

with postmenopausal bleeding for 1 week, having been menopausal since age 53. An endometrial biopsy revealed endometrial carcinoma. The patient was unaware that she had a genital tract anomaly. Her obstetric history consisted of 3 spontaneous vaginal deliveries at term with no complications. She underwent a pelvic transabdominal sonography that revealed the presence of a presumed bicornuate or didelphys uterus with two endometrial cavities. The thickness of the endometrial images was 6 mm and 18 mm in the right and left horns, respectively.

Total abdominal hysterectomy, bilateral salpingo-oophorectomy, and staging (bilateral pelvic and paraaortic lymph node dissection, omentectomy, and pelvic washings) were performed. At the time of surgery, it was determined that the patient had a bicornuate uterus (Fig. 1). Macroscopic examination revealed a bicornuate uterus (left horn 50×25×30



Fig. 1. Hysterectomy specimen showing uterus bicornis unicollis.

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Fig. 2. Pathology specimen showing left horn with obvious polypoidal tumour and apparently normal right horn.

mm; the right horn 40×35×15 mm) with a single cervix, measuring 35×30×25 mm. The attached bilateral tubes and ovaries were normal. The uterus revealed an irregular polypoidal mass measuring 3×25×5 mm in the fundus and body of the left horn (Fig. 2).

Pathologic review confirmed the presence of a well-differentiated endometrioid adenocarcinoma of the endometrium, arising within the left horn of the bicornuate uterus with no definite myometrial invasion. The right uterine horn was lined with normal endometrium. Pelvic and paraaortic lymph nodes, peritoneal washings, omentum and bilateral ovaries were negative for metastases. No adjuvant therapy was administered, and at the time of this writing she was asymptomatic and without evidence of disease. The disease was consistent with FIGO stage IA.

DISCUSSION

Müllerian anomalies have not been implicated as a significant risk factor for the development of cervical, uterine and ovarian cancers; however several reports describe the existence of various tumours in anomalous uteri.⁶ A correlation between the presence of uterine malformations and the incidence of endometrial cancer has never been reported, and in the present literature, there are only few reports of endometrial cancer arising in patients with müllerian abnormalities.

This case illustrates the importance of existence of uterine abnormalities in the differential diagnosis when evaluating postmenopausal bleeding. The prevalence of these anomalies may be higher than reported due to the asymptomatic nature of some of these cases, as was noted in our case, a woman who had three pregnancies and was 58 years old when the uterine anomaly was diagnosed secondary to the diagnosis of carcinoma. If only the right uterine horn had been biopsied, there would have been a delay in the diagnosis of endometrial adeno-

carcinoma. Furthermore, many of the described endometrial carcinomas of bicornuate uteri existed in only one horn of the uterus, implying that there is a 50% chance of obtaining a biopsy of the benign horn at the time of initial workup, thereby delaying treatment.^{7,8} In the presence of two uterine horns, a bilateral endometrial biopsy should be performed in order to reduce the risk of delayed and/or inadequate diagnosis. Physicians should remember the possible existence of a separate uterine cavity when endometrial cancer is clinically suspected but histology fails to confirm the diagnosis. This emphasizes the importance of a careful physical examination and/or radiographic evaluation if required. Recent studies have documented the utility of sonographic and magnetic resonance imaging in delineating the shape of the uterus as well as the cervical, vaginal and adnexal components.⁹

It is unclear why the cancer developed in only one uterine horn while the endometrium in the contralateral horn remained unaffected, as in our case with a bicornuate uterus. Invasive endometrial adenocarcinomas, however, are frequently a focal process surrounded by benign endometrium. A review of the literature revealed no apparent predilection for malignant transformation in the right versus left uterine horn. The diagnosis of uterine malformations may be overlooked because the malformations are silent. In such cases, a high index of suspicion is necessary to avoid diagnostic delay. Our patient was not aware of her anatomic abnormalities.

The treatment of endometrial cancer in patients with bicornuate uterus is the same as with single uterus but the management of this tumor is made more difficult by the presence of two horns. Each horn may be affected independently by the carcinoma, it may affect one or both horns and various histological types of endometrial carcinoma may be present in each horn. Finally, the overall survival rate in anomalous uteri appears to depend on stage, nodal involvement, histological grade and depth of penetration.

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