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Endometrial cancer in unicornuate uterus: a case report

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Summary

Purpose of Investigation: Müllerian anomalies have not been implicated as a significant risk factor for the development of cervical, uterine, and ovarian cancers; in the present literature, there are only a few reports of endometrial cancer arising in patients with Müllerian abnormalities. To the best of the authors' knowledge, this is the first reported case of endometrial cancer arising in a patient with unicornuate uterus. *Case Report:* A 69-year-old Caucasian woman underwent clinical examination and office hysteroscopy with endometrial biopsy because of abnormal post-menopausal bleeding. The diagnosis was endometrial cancer in unicornuate uterus, hence the patient underwent total hysterectomy with pelvic lymphadenectomy. *Conclusion:* Uterine malformations and genetic disorders may cause a delayed diagnosis of gynaecological cancers. Gynaecological examination in asymptomatic patients and differential diagnosis in abnormal uterine bleeding patients should be considered.

Key words: Endometrial cancer; Unicornuate uterus; Diagnosis; Therapy.

Introduction

Uterine Müllerian malformations represent a heterogeneous group of congenital anomalies resulting from the underdevelopment of the Müllerian ducts, disorders in their fusion, and/or alterations in septum reabsorption. Prevalence of uterine malformations is estimated to occur in 0.4% (0.1–3%) of the general population [1, 2].

The 1988 classification of Müllerian malformation by the American Fertility Society (AFS) defines the unicornuate uterus as a condition where the uterus is formed from only one of the paired Müllerian ducts, while the other duct does not develop or only in a rudimentary fashion [3, 4]. Two main techniques for the diagnosis of these malformations combine the study of the uterine fundus and cavity: magnetic resonance imaging (MRI) and three-dimensional (3D) ultrasound [5, 6].

Uterine Müllerian malformations are related to many gynecological diseases, in particular to infertility and recurrent miscarriage [7], while a small number of evidences of the association between uterine malformation, such as unicornuate uterus, and genital tumors exist [8-16]. Therefore, a few documented cases of cervical carcinoma arising in unicornuate uteri have been described [8], whereas endometrial malignancy occurring in conjunction with this Müllerian anomaly has not been reported.

In this paper the authors report the case of an endometrial cancer in a unicornuate uterus.

Case Report

A 69-year-old, Caucasian woman came at our attention in February 2013 at the Department of Gynaecology, Obstetrics and Reproductive Science of the Second University of Naples because of a post-menopausal abnormal uterine bleeding occurring a few months earlier. She had been in menopause for about 20 years and had never undergone surgery. She had two spontaneous deliveries and one spontaneous miscarriage. The woman was affected by HCV-related liver disease.

Clinical examination revealed healthy cervix and vagina. Uterus was mobile and deflected to the right. No adnexal masses were palpable. The woman underwent transvaginal ultrasonography, which showed an irregularly formed uterus with almost normal size for her age (52×35×46 mm), but the endometrium was thickened: 11 mm.

Subsequently, the woman underwent office hysteroscopy with endometrial biopsy, with a pathological report of endometrial adenocarcinoma, endometrioid type. MRI was performed to evaluate myometrial infiltration and abdominal diffusion and showed a unicornuate uterus constituted only by its right side (Figure 1). The examination indicated a marked myometrial infiltration at the level of the fundus. The other pelvic organs did not appear to be affected by cancer infiltration. No iliac or retroperitoneal lymphadenopathy was detected. Thus, the woman underwent abdominal total hysterectomy, bilateral adnexectomy, bilateral pelvic lymph node dissection, peritoneal washing, and removal of a peritoneal lesion. At the time of surgery, it was determined that the patient had a unicornuate uterus, with the left side of the uterus completely absent.

Macroscopic examination revealed a unicornuate uterus (size: 9×4.5×2.5 cm) with diffusely thickened endometrium for the presence of a vegetating lesion occupying almost the entire uterine cavity; right ovary was sized 3×2×1.5 cm and right fallopian tube

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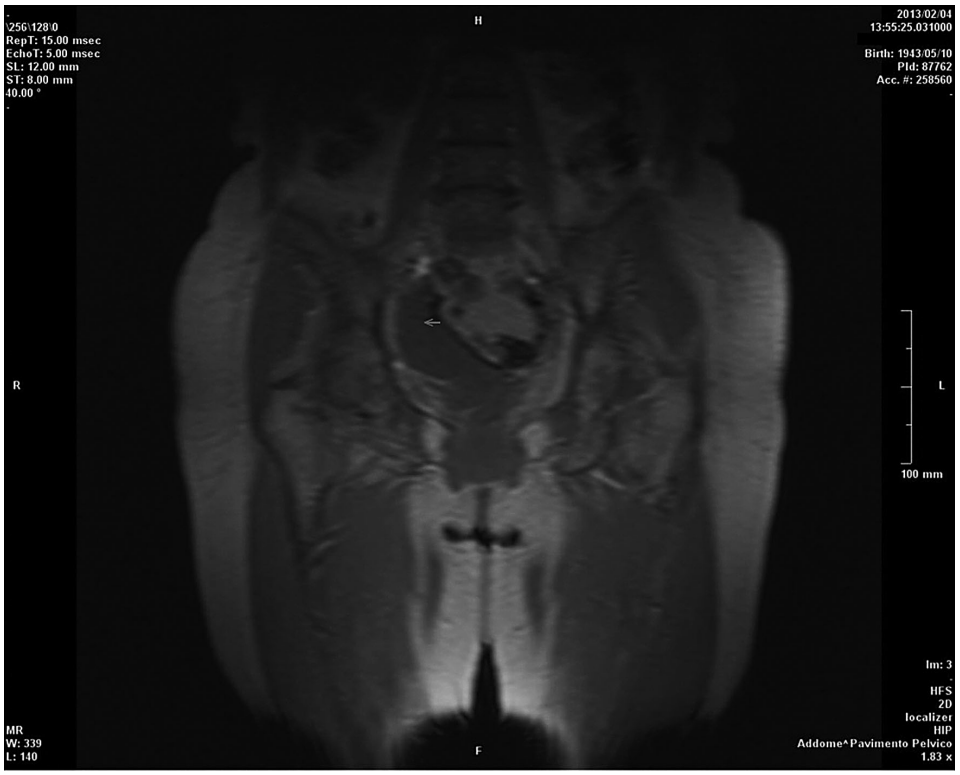

Figure 1. — MRI image showing a unicornuate uterus constituted only by its right side. The endometrium is thickened because of the presence of the endometrial cancer (arrow).

long five cm; left ovary was sized 3×2×1 cm with fallopian tube long four cm.

Microscopic examination revealed a well-differentiated (G1) endometrioid adenocarcinoma of the endometrium, infiltrating the myometrium for more than a half. The examination also revealed the presence of neoplastic vascular emboli. Cervix, adnexa, and peritoneal fluid resulted free from disease. The peritoneal lesion revealed to be a calcific concretion. Twenty-one lymph nodes were examined and all of them resulted negative for malignancy. Cancer Stage was FIGO IB. After discharge and an oncological consultancy, the patient is now undergoing a standard follow-up.

Discussion

As a small number of evidences of the association between uterine malformation and genital tumors exist [8-16], Müllerian anomalies have not been implicated as a significant risk factor for the development of cervical, endometrial, and ovarian cancers. Therefore, endometrial malignancy, which is the most common malignant tumor of the female genital tract in developed countries [17], occurring in conjunction with unicornuate uterus has not been reported.

At present, a few reports of endometrial cancer arising in patients with Müllerian abnormalities exist. In particular, six cases of endometrial cancer in didelphys uterus [10-13] and three cases of endometrial cancer in bicornuate uterus [14-16] have been described.

To the best of the present authors' knowledge, this report represents the first case of endometrial cancer developing in a unicornuate uterus, whereas some cases of cervical and ovarian cancers arisen in unicornuate uterus have been described [8, 9].

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 the present case, in a woman who had three pregnancies and was 69-years-old when the uterine anomaly was diagnosed secondary to the diagnosis of carcinoma. This emphasizes the importance of a careful physical examination, radiographic evaluation, and/or sonographic and MRI if required [18].

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