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Case Report

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Uterus Didelphys with Septate Cervix and Unilateral Endometrial Carcinoma: A Case Report

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Abstract

Uterine malformations owing to defects of fussion of müllerian ducts are not rare, but cases of complete duplicity (didelphys or bicornuate-bicollis uterus) with double vagina are rarer and usually have two cervices, one connecting each vagina. However, the combination of a uterus didelphys in the superior uterine segment with a septate cervix is either uncommon or not well diagnosed or reported. This case report describes a 74-year-old woman who had given birth several times and now exhibits uterus didelphys with an endometrial carcinoma in one of the hemiuteri; there was no pathology in the other hemiuterus. The cervical portion was single but septate, and the vaginal septum had been previously removed (resorption defect). The transvaginal ultrasonography identifying the malformation, the endometrial biopsy of both sides in the presence of indicative clinical symptoms and the hysteroscopy are fundamental for diagnosis.

Keywords: Uterus didelphys; Septate cervix; Unilateral endometrial carcinoma; Genital malformation

Introduction

The combination of a bicornuate uterus (or didelphys in the superior uterine segment) with a septate cervix, although logically a transitional type, is either quite rare or not well diagnosed or reported. This case report describes a 74 year-old woman, gravid 8, para 7, who now exhibits uterus didelphys with an endometrial carcinoma in one of the hemiuteri; there was no pathology in the other hemiuterus.

Case Report

The patient was 74 years of age and became menopausal at 51. She had 8 previous pregnancies: 1 abortion and 7 vaginal births (forceps delivery in some), with 4 offspring still living. She presented with diabetes, obesity (weighing 88 kg, 1.51 cm tall) and hypertension (170/90 mmHg). A consultation in January 2005 was ordered for scarce metrorrhagia that had been occurring the previous 3-4 months. Physical examination and hysteroscopy proved inconclusive at that time, but uterine malformation and pathology on one of the two sides of the uterus were suggested. The vagina and cervix were both normal, as well as the cervical smear. The patient stated that during her deliveries, some of the vaginal septum was resected. The transvaginal ultrasonography (TVU) showed bicornuate uterus, endometrial atrophy in the right hemiuterus and irregular thickening of 1.5 cm, suggesting carcinoma in the left hemiuterus (Figure 1A). Endometrial biopsies were taken from both sides; the histopathological study showed acellular mucoid content on the right side and moderately differentiated endometrioid adenocarcinoma on the left side. Meanwhile, the hysteroscopy was repeated, which revealed the images shown in figure 1B. She had a septate cervical channel and a normal endometrial cavity in the right hemiuterus, with atrophy. In the left hemiuterus, mammillated irregularities with increased vascularisation, indicative of endometrial carcinoma, were observed.

After completing the pre-operative studies, the patient underwent laparotomy with total hysterectomy, bilateral salpingooophorectomy and pelvic lymph node sampling in March 2005. The surgical specimen from the hysterectomy is shown in figures 1C-1E. The patient had uterus didelphys in the superior uterine segment with a single cervical portion, which, as the section shows, leads to a septate cervix. The right hemiuterus had no endometrial pathology, but there

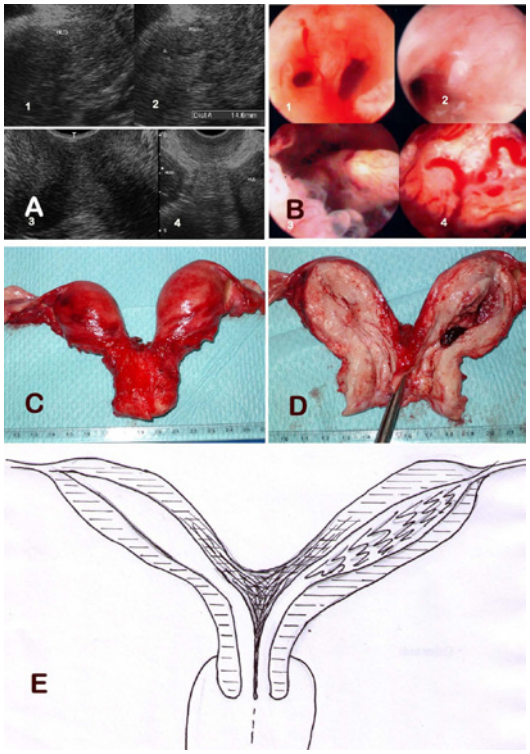

Figure 1: A) Transvaginal ultrasonography images. 1 and 2, longitudinal cross sections of right and left hemiuterus. 3 and 4, transversal cross sections, at cervical portion in 3, at corpus of both hemiuteri in 4. **B)** Hysteroscopical images. 1, endocervix; 2, right hemiuterus; 3 and 4, left hemiuterus showing adenocarcinoma images. **C)** Surgical specimen from hysterectomy. **D)** Longitudinal section of the anterior wall of both hemiuteri. **E)** A graphical representation of the uterine malformation.

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was evidence of endometrial carcinoma occupying the entire uterine cavity in the left hemiuterus.

The histopathological study confirmed well-differentiated endometrial carcinoma in the left hemiuterus, FIGO IB stage; the lymph nodes were negative and cytology from the peritoneal lavage was negative for malignancy. No subsequent adjuvant treatment was performed. After 7 years of follow-up our patient is in good clinical condition without any evidence of disease.

Comments

The case presented here was one of uterus didelphys in the superior uterine segment (a defect of fusion) with a septate cervix and vagina that had been previously removed (defect from reabsorption). This is an isolated Müllerian anomaly (group 3 of the Acien classification [1]) of transitional type with divergence in the processes of fusion and reabsorption of the superior (convergent) and inferior (divergent) uterine segments [2]. However, the most interesting issues of this case are: 1. the obstetric history of the patient, including 7 vaginal births in spite of the uterine malformation; and 2. the development of unilateral endometrial carcinoma.

In spite of presenting general risk factors (diabetes, obesity, hypertension), the carcinoma had developed throughout the left hemiuterus cavity without any effect on the right side whatsoever. There are several cases published describing unilateral carcinoma in septate uteri [3,4] and in uterus didelphys or bicornuate [5-8], although the most frequent manifestation affects both uterine horns. The TVU identifying the malformation, the endometrial biopsy of both sides in the presence of indicative clinical symptoms and the hysteroscopy are fundamental for diagnosis.

It would be useful to investigate probable correlations between specific factors and endometrial cancer risk associated to uterus malformations. This may improve diagnostic procedure and treatment.

Disclosure of Interests

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this paper. The authors declare that they have no competing interests

Contribution of Authorship

MMB reviewed the case, participated in its study and surgical operation and reviewed the manuscript. JG participated in its study, made the hysteroscopies and reviewed the manuscript. PA studied the patient, designed the study, reviewed the literature, made the figures and wrote the paper. PA had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Details of Ethics Approval

Not required for case studies in Spain. Written informed consent was obtained from the patient for publication of case and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this Journal.

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