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Original Article

Acquired uterine vascular abnormalities associated with persistent human chorionic gonadotropin: Experience at a Korean teaching hospital

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ABSTRACT

Objective: The aim of this study was to describe our experience with the diagnosis and management of acquired uterine vascular abnormalities associated with persistent human chorionic gonadotropin (hCG). Through this case series, we sought to establish our protocol for the treatment and follow-up of uterine vascular lesions associated with persistent hCG.

Materials and methods: We examined the clinical presentations of 28 Korean women with acquired vascular uterine abnormalities associated with persistent hCG who were seen in the Department of Obstetrics and Gynecology of the Gangneung Asan Teaching Hospital, Gangneung-si, Korea between October 2006 and July 2012 and retrospectively reviewed their medical records.

Results: The mean patient age was 32.5 ± 6.4 years, and the mean parity was 1.4 ± 1.2. The mean size of the vascular lesions in color Doppler sonography and multidetector computed tomography with angiography was 3.1 ± 1.6 cm and 3.9 ± 1.6 cm, respectively. Multidetector computed tomography revealed arteriovenous malformation-like vascular lesions ($n = 15$) and pseudoaneurysms ($n = 3$). Treatments included clinical observation ($n = 11$), uterine artery embolization ($n = 11$), hysterectomy ($n = 4$), and chemotherapy, including single methotrexate (MTX) treatment and combination chemotherapy ($n = 9$).

Conclusion: When the uterine vascular lesion is not decreased, or if weekly clinical follow-up reveals that the serum β-hCG level is persistently elevated or sustained in conjunction with vaginal hemorrhage, a proper management strategy is required.

Introduction

Uterine vascular abnormalities are rare clinical findings. Consequently, only a few cases of uterine vascular abnormalities have been reported in the literature. Because these vascular lesions were often overlooked by obstetricians and gynecologists and a standard protocol for diagnosis and therapy has not yet been established, there are difficulties associated with the diagnosis and treatment of uterine vascular lesions [1].

Vascular abnormalities that affect the uterine arteries include congenital arteriovenous malformations (AVMs), acquired AVMs, arteriovenous fistulas, true aneurysms, and pseudoaneurysms [2]. Congenital uterine AVMs are rare lesions resulting from the abnormal embryonic development of primitive vascular structures that cause abnormal communications between arteries and veins [3]. We have encountered no cases of congenital AVM at our institution. Congenital AVMs usually have multiple feeding arteries, a tangle of vessels with histological characteristics of both arteries and veins, and many large draining veins [4]. Acquired uterine AVMs typically result from trauma, such as a prior dilation and curettage, a therapeutic abortion, uterine surgery, or direct uterine trauma [5,6]. Endometrial carcinoma, cervical carcinoma, and gestational trophoblastic diseases are other possible causes of acquired uterine AVMs [7,8]. If a hypervascular lesion with typical early venous filling within the myometrium is accompanied by a negative serum β-hCG level, a uterine AVM diagnosis should be considered [9]. However, uterine hypervascular lesions with turbulent flow have been observed with an elevated serum β-hCG level in patients with gestational trophoblastic disease (GTD) and

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retained products of conception (RPOC) [10]. Uterine vascular abnormalities with elevated serum β-hCG levels may be distinguished from acquired AVM and associated with GTD and RPOC. However, there are some difficulties associated with the pathologic diagnoses of these lesions due to the limited availability of surgical specimens. Therefore, the lesions with elevated serum β-hCG could be considered to be acquired uterine vascular abnormalities associated with persistent hCG and distinguished from AVM.

The most important clinical manifestation of uterine vascular abnormalities is recurrent and/or profuse vaginal bleeding, which may result in hypovolemic shock and/or death. Vaginal bleeding arising from these vascular abnormalities may be aggravated by dilation and curettage performed for the purpose of biopsy and bleeding control, unlike excessive uterine bleeding due to other common causes [6].

The management of these vascular lesions ranges from a conservative approach, such as observation or medication, to invasive procedures, such as uterine artery embolization or emergency hysterectomy. However, most reports in the literature consist of only a few cases, and experience with vascular abnormalities at individual institutions is limited. In this study, we present the image patterns of uterine vascular abnormalities associated with persistent hCG using sonography with color Doppler and multidetector computed tomography with angiography and describe our findings regarding the immediate results and long-term outcomes of these lesions. By examining this case series, we sought to establish our protocol for the treatment and follow-up of uterine vascular lesions associated with persistent hCG.

Materials and methods

This study was approved by Gangneung Asan Hospital Institutional Research Ethics Committee (2010-056). We reviewed the medical records of patients with acquired uterine vascular abnormalities associated with persistent hCG diagnosed and treated at Gangneung Asan Hospital, Gangneung-si, Korea between October 2006 and July 2012. During the study period, a total of 3947 pregnancies, including abnormal pregnancies, were diagnosed at our outpatient department. At our institution, uterine vascular abnormalities associated with persistent hCG were diagnosed in 28 patients (0.71%). In this case series, patients with intrauterine vascular abnormalities with elevated serum β-hCG measurements were enrolled. According to our study design, patients with normal intrauterine pregnancy or ectopic pregnancy, in addition to those with uterine vascular lesions associated with negative serum β-hCG measurements, would be excluded from the case series because serum β-hCG measurements are elevated or sustained in normal intrauterine pregnancy and ectopic pregnancy regardless of the presence of uterine vascular abnormalities. Additionally, in cases of normal intrauterine pregnancy, multidetector computed tomography with angiography should be avoided due to the radiation hazard associated with this technique. For these reasons, we planned to exclude the cases of normal intrauterine pregnancy with uterine vascular abnormalities. However, there were no cases of uterine vascular abnormalities with normal intrauterine pregnancy or ectopic pregnancy.

When patients with vaginal spotting or bleeding and whose vital signs were stable visited our outpatient division, physical examinations including a vaginal examination, urine hCG test, and pelvic sonography with color Doppler were performed to detect uterine abnormalities. Three cases without symptoms were observed by chance during routine gynecological examinations. For suspected uterine vascular lesions, we performed serial serum β-hCG measurements and transvaginal sonography of the intrauterine cavity to rule out a normal intrauterine pregnancy or an ectopic pregnancy by additional image evaluation. In patients with stable vital signs, multidetector computed tomography with angiography was performed to evaluate the uterine vascular lesions.

The management options for these uterine vascular abnormalities consisted of observation, single chemotherapy with methotrexate (MTX), combination chemotherapy (EMA or EMA-CO), transcatheter uterine artery embolization, and hysterectomy. At our institution, the protocol for the treatment and follow-up of vascular abnormalities was established based on clinical case series and case reports in the literature (Figure 1). Based on our protocol for treatment and follow-up, if there was profuse vaginal hemorrhage with stable vital signs, transcatheter uterine artery embolization was performed. If the patients had unstable vital signs because of profuse vaginal bleeding, an emergency hysterectomy was performed. In cases with vaginal spotting or no specific symptoms, clinical follow-up, including transvaginal sonography with color Doppler and serum β-hCG measurements, was performed weekly. For cases in which two or three consecutive clinical follow-ups demonstrated a sustained or elevated level of serum β-hCG, chemotherapy was considered. For cases in which the size of the uterine vascular lesions increased or failed to regress during clinical follow-up, a transcatheter uterine artery embolization was performed. After treatment, we performed periodic physical examinations and measured the patients' serum β-hCG levels in the outpatient division to check for the recurrence of uterine vascular abnormalities.

Data are expressed as mean ± standard deviation (SD) unless stated otherwise. All statistical analyses were performed with SPSS version 13.0 for Windows (SPSS Inc., Chicago, IL, USA).

Results

The mean patient age was 32.5 ± 6.4 years (range, 19–42 years), and the mean parity was 1.4 ± 1.2 (range, 0–4). Twenty-five patients had vaginal hemorrhage, two patients had no specific symptoms, and one patient had amenorrhea. Regarding surgical history, nine patients had undergone a cesarean section one or more times, one patient had undergone an appendectomy, and one patient had undergone laparoscopy.

The antecedent pregnancy events of the patients were the following: missed abortion ($n = 11$), intrauterine curettage for artificial abortion ($n = 8$), hydatidiform mole ($n = 5$), cesarean scar pregnancy ($n = 2$), and unknown event ($n = 2$). With these antecedent pregnancy events, the patients had undergone surgical procedures such as dilation and curettage ($n = 18$), suction and curettage ($n = 5$), no procedure ($n = 3$), MTX injection ($n = 1$), and unknown ($n = 1$) before acquired uterine vascular abnormality was diagnosed. In two patients, uterine vascular lesions were diagnosed during a missed abortion (Figure 2).

The interval from the antecedent pregnancy event to the diagnosis of an acquired uterine vascular lesion ranged from 0 days to 192 days (mean ± SD: 48.9 ± 41.5 days). All patients had an elevated serum β-hCG level at the time of diagnosis, and the mean serum β-hCG level was 4,136.0 ± 10,399.9 mIU/mL, with a range from 3.9 mIU/mL to 48,427.0 mIU/mL. Therefore, we classified these lesions as acquired uterine vascular abnormalities associated with pregnancy because the serum β-hCG level is not elevated in uterine AVMs. A gray-scale transvaginal sonography with color Doppler scan was performed in 27 patients, and one patient underwent an emergency hysterectomy without a color Doppler scan because of unstable vital signs. Color Doppler sonography of the vascular lesions revealed a high-flow vascular lesion or tangles of vessels with multidirectional high velocity flow producing a color mosaic pattern. In 22 of 27 patients, multidetector computed tomography with angiography was performed. In five cases, uterine vascular lesions were diagnosed with color Doppler sonography without multidetector computed tomography.