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BRIEF COMMUNICATION

Hematometra Due to Cervical Stenosis in a Postmenopausal Woman with Incidental Ovarian Steroid Cell Tumor: A Case Report

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Vaginal bleeding is reported among 4%–11% of postmenopausal women. Hematometra is commonly associated with cervical stenosis due to senile atrophy, radiotherapy, or a uterine neoplastic lesion in women of postmenopausal age. Ovarian steroid cell tumor is a rare hormone-secreting tumor subtype accounting for approximately 0.1% of all ovarian tumors. Here we report a case of hematometra in a postmenopausal woman with high estrogen levels who was later diagnosed with a steroid cell tumor.

Key Words: Hematometra, Menopause, Ovarian neoplasm

INTRODUCTION

In postmenopausal women, hematometra is commonly associated with cervical stenosis due to senile atrophy, radiotherapy, or neoplastic lesions involving the uterus [1]. Ovarian steroid cell tumors are a rare subtype of hormone-secreting tumors that account for approximately 0.1% of all ovarian tumors [2]. Here, we report a case of hematometra in a postmenopausal woman with high estrogen levels who was later confirmed to have a steroid cell tumor.

CASE REPORT

A 56-year-old multiparous woman (gravida 3, para 3) visited the emergency room of Asan medical center because of vaginal bleeding with progression to cramping abdominal pain. She was menopausal since the age of 49 years and had never received hormone replacement therapy. The patient was 158 centimeters tall and weighed 55 kilograms. On vaginal ultrasonography, 57 × 35 × 34 mm of hematometra mainly located in the right upper cervix was identified (Fig. 1A). Eight years prior, the patient was diagnosed with cervical carcinoma in situ and she underwent conization. A Pap smear was performed annually at the same hospital, and hematometra was not observed on ultrasound 1 year before the onset of symptoms (Fig. 1B). The hematometra supposedly developed from occlusion as a result of cervical manipulation. However, the cause of the endometrial bleeding remained unclear. The patient had underlying Henoch-Schoenlein purpura with nephrotic syndrome and was undergoing peritoneal dialysis. Additionally, she was diagnosed with breast cancer but showed no recurrence after treatment.

For the treatment plan, the patient visited the outpatient clinic, and further work-up was performed. Detailed ultrasonography revealed a stationary hematometra (56 × 34 × 35 mm) and an 18-mm left ovarian mass. The ovarian mass had an internal echo without a solid portion and vascularity (Fig. 2A). The blood test revealed a serum follicle-stimulating hormone level of 165 U/mL, indicating menopause status, and an estradiol level of 91.4 pg/mL (normal range in postmeno-

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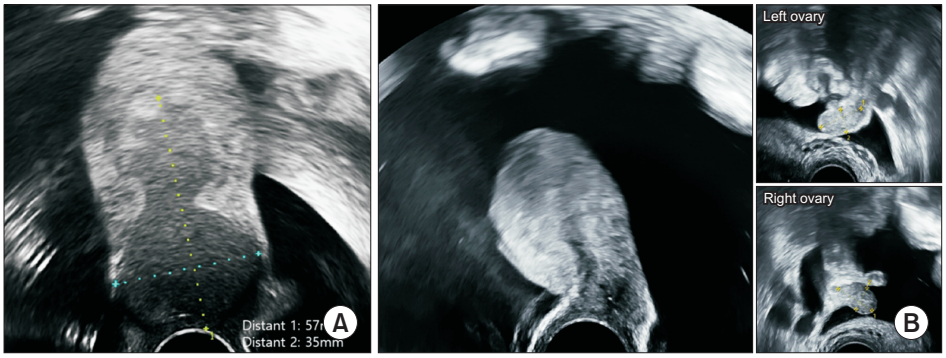

Fig. 1. The vaginal ultrasonography findings. (A) Hematometra sized 57 × 35 × 34 mm. (B) Previous normal uterus and ovaries. The patient was on peritoneal dialysis. Therefore, fluid collection in the pelvic cavity was present.

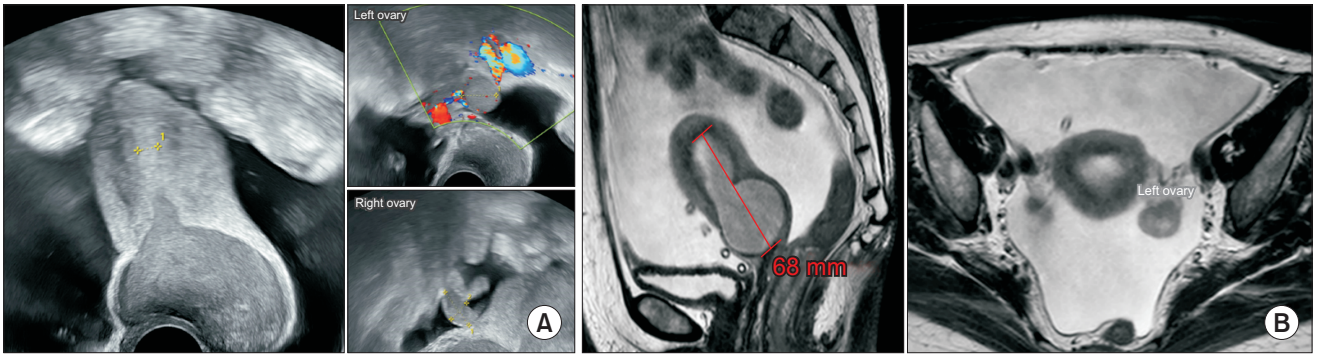

Fig. 2. Follow up vaginal ultrasonography and magnetic resonance image (MRI). (A) Hematometra was stationary but the left ovary mass was identified. The ovarian mass had an internal echo without a solid portion and vascularity. (B) Only hematometra (size, 68 mm) was identified without endometrial malignancy. Only the left ovary was visible, and the MRI report revealed no abnormality in the left ovary.

pausal women: less than 10 pg/mL) [3], which was much higher than that expected for a postmenopausal woman. The other laboratory results were nonspecific. An endometrial biopsy was performed for suspected endometrial cancer. However, the cervix was completely occluded; therefore, biopsy could not be performed. On magnetic resonance imaging (MRI), only hematometra was identified, without endometrial malignancy. The MRI report revealed only the left ovary without any abnormality (Fig. 2B).

Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed without any complications. A slight increase in the uterus size was identified, and a 10 × 10 × 10 mm mass with a smooth surface was noted in the left ovary (Fig. 3). The final pathology result of the left ovary was a multifocal steroid cell tumor associated with Leydig cell hyperplasia. The notable tumor foci measured 16 × 13 × 13 mm and 3 × 3 × 3 mm, with no mitotic activity, no necrosis, and no significant nuclear atypia. The endometrium was confirmed to be negative for malignancy. The patient recovered and was discharged.

DISCUSSION

Vaginal bleeding is reported among 4%–11% of postmenopausal women [4]. It occurs mainly due to an atrophic endometrium and vagina owing to insufficient estrogen. However, the development of hematometra in postmenopausal women is rare. Generally, cervical stenosis is preceded by senile atrophy, radiotherapy, or neoplastic lesions involving the uterus that lead to development of hematometra [5]. In this case, cervical stenosis was caused by previous conization. However, the cause of a relatively large amount of hematometra was not explained from the history of conization.

Chronic kidney disease (CKD) causes hormonal imbalance and menstrual disorders [6]. Estradiol, progesterone, and follicle-stimulating hormone levels are similar among premenopausal women with CKD. However, an absence of sex hormone level cyclicity in women with CKD has been reported. According to previous studies, serum estrogen levels are low in postmenopausal women with CKD treated with hemodialysis [7]. Such hormonal imbalance in patients with

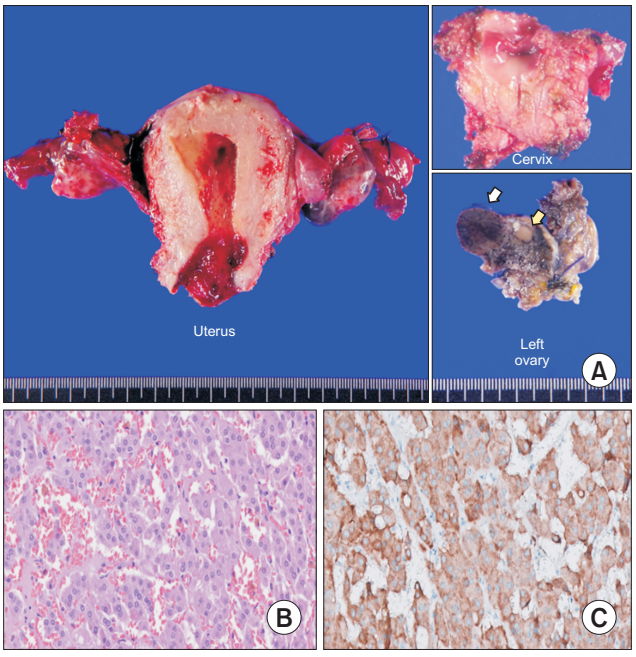

Fig. 3. The pathologic findings of the case. (A) Uterus, cervix, and left ovary. A cross-section of the left ovary demonstrated the solid yellow to brown colored tumor. The black and yellow arrows indicate steroid cell tumors. (B) Microscopically, the tumor was composed of granular eosinophilic and vacuolated cytoplasm, which led to the diagnosis of a steroid cell tumor (stain, hematoxylin and eosin; magnification, × 40). (C) The steroid cells positive for inhibin are demonstrated in brown color (stain, inhibin; magnification, × 40).

CKD causes bleeding in the endometrium, but there may also be an imbalance due to hormones secreted by tumors [2]. This patient had an incidental ovarian steroid cell tumor that produced hormones.

An ovarian steroid cell tumor is rare and it mainly produces androgen hormones, resulting in virilization. According to a previous study, steroid cell tumors account for < 0.1% of all ovarian tumors and the mean age of presentation is 43 years [8]. The clinical symptoms of steroid cell tumors vary depending on the hormone secretion. Hyperandrogenism is commonly found among patients with steroid cell tumor, occurring in up to 50% of patients. Precocious puberty can also occur in children. In adults, virilizing signs such as hoarseness of voice, hirsutism, and amenorrhea have been reported [2]. In this case, signs of virilization including acne, hirsutism, and male pattern alopecia were not identified.

In a previous report, hyperestrogenism caused by ovarian steroid cell tumors was reported in 6%–23% of patients [8]. In this case, the patient also showed elevated estradiol levels despite menopause. The consequences of hyperestrogenism in steroid cell tumors include postmenopausal bleeding, menorrhagia, and endometrial carcinoma. In this case, the endometrial bleeding could be attributed to hyperestrogenism caused by the ovarian steroid cell tumor.

The negative feedback of hypothalamic, pituitary and ovarian (H-P-O) axis remains intact in postmenopausal women [9]. Therefore, when postmenopausal women with high follicle stimulating hormone (FSH) levels are treated with estrogen, FSH levels are reduced; however, they do not return to premenopausal levels [10]. The present patient showed high FSH levels (165 mIU/mL) despite the elevated concentration of serum estradiol (91.4 pg/mL). Rarely, high serum estradiol levels are associated with estradiol-producing tumors in postmenopausal women. This patient's high estrogen levels could be related to the estrogen-producing steroid tumor; 4 years ago, this patient's estradiol levels were as low as 12.4 pg/mL (FSH levels were 97.7 mIU/mL), and there were no specific findings in the bilateral adnexa as per the abdominal pelvis computed tomography performed at that time. Further, the possibility of false-positive results cannot be excluded. However, a case report revealed high estradiol levels without suggesting a tumor status even after bilateral adnexectomy [11]; the other sex steroids hormones including FSH were at normal levels of a postmenopausal woman. The false positive result was normalized after immunoassay with sheep-derived monoclonal antibodies, which implied that the patient had a cross reaction with irregular antibodies. CKD could also be attributed to our patient's abnormal hormone profiles; altered or dysregulated estrogen and estrogen receptor signaling pathways were observed in CKD, which itself is known to be associated with H-P-O dysfunction [12]. Additionally, a recent study revealed that higher FSH levels were associated with lower glomerular filtration rate in postmenopausal women [13]. However, the relationship among estrogen, FSH, and renal disease remains unknown. The cause of the abnormal negative feedback of the H-P-O axis could not be determined in this patient. Nevertheless, it is necessary to monitor the hormone level periodically after surgery.

The treatment of steroid cell tumors involves surgical removal of the tumor. In young patients, excision of the primary lesion by unilateral oophorectomy can be considered because the frequency of bilateral occurrence is only 6%. However, hysterectomy with complete

surgical staging is recommended when fertility is not considered [14]. Malignant ovarian steroid cell tumors have also been reported [15]. However, the histological criteria for benign and malignant steroid cell tumors are not yet determined. The following pathologic features are related to malignancy: tumor size > 7 cm, significant mitotic activity, necrosis, hemorrhage, and significant nuclear atypia. According to the pathological report, the patient had no malignant features.

In conclusion, we report a rare case of hematometra in a postmenopausal woman with a hormone-producing ovarian steroid cell tumor. In case of vaginal bleeding or abdominal pain with a large amount of hematometra in postmenopausal women, anatomical abnormalities, endocrine abnormalities, and ovarian origin should be considered.

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