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Perimenopausal Amenorrhea with Cervical Atresia and Hematotrachelos

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Obstruction of the lower female genital tract leading to proximal dilatation and development of hematotrachelos is most commonly a result of congenital abnormalities such as an imperforate hymen, a complete transverse vaginal septum, or rarely, cervical atresia. Acquired hematotrachelos is rare, and the possible etiology includes iatrogenic trauma to the uterocervix such as cervical surgery, dilation and curettage, obstetric laceration, cervical or endometrial carcinoma, and radiation therapy. We report an unusual case of a patient in who spontaneously acquired hematotrachelos developed in the perimenopausal period. Combined speculum inspection of the cervix and recognition of transvaginal echographic features assisted the diagnosis. In addition, we tried a new treatment method, using the firm plastic end of an Ipas Karman cannula (6 mm diameter and 6 cm length) with nylon fixation retained for 1 month that resulted in a patent cervical canal.

Key words: hematotrachelos, secondary amenorrhea, perimenopause

INTRODUCTION

Obstruction of the female genital tract results in menstrual blood accumulating in the vagina (hematocolpos), uterine cervix (hematotrachelos), uterine cavity (hematometra), or fallopian tubes (hematosalpinx) and is most commonly a result of congenital abnormalities such as an imperforate hymen, a complete transverse vaginal septum, or rarely, cervical atresia.[1-3] These entities generally present at puberty when symptoms arise from the retention of menstrual blood or from bladder outlet obstruction secondary to compression of the urethra.[4,5] Acquired obstruction of the lower female genital tract is rare and is usually caused by iatrogenic trauma, obstetric laceration, or cervical or endometrial carcinoma.[6,7]

Acquired obstruction of the lower female genital tract caused by the spontaneous cervical atresia might be resulted from the low level of estrogen in perimenopausal period. Perimenopause is a period during which women experience physiological changes and begin menopause transition. It's typically several years before the natural menopause, which means no menstruation for 12 consecutive months. In perimenopause years, hormones produced by the aging ovaries fluctuate and lead to irregular menstrual patterns including irregular length of the periods, the intervals, and the level of flow. Here we describe a rare case of secondary amenorrhea in the perimenopausal period caused by spontaneous cervical atresia with isolated hematotrachelos mimicking an ovarian tumor. We discuss the key diagnostic points and an appropriate treatment method

CASE REPORT

A 48-year-old female, gravida 2 para 0, presented with lower abdominal pain, overdue period and last menstrual period 6 months previously. She was referred from the local clinic because of suspicion of an ovarian tumor and menopause. Her medical history was unremarkable and her previous gynecological history showed 2 elective dilatation and curettage procedures for abortion at the ages of 29 and 36, and an exploratory laparotomy for ovarian cyst at the age of 36. She had regular menstruation without history of metrorrhagia, menorrhagia, and dysmenorrhea. Not until 1 year prior to this admission did she begin to experience hypomenorrhea following extracyclic vaginal spotting. Although amenorrhea developed over 6 months, she did not receive adequate treatment until an ovarian tumor as large as 6 cm was informed.

Physical examination showed a healthy, nonobese, nonhirsute female of height 155 cm, weight 50 kg and thus body mass index of 20.8. The abdomen was soft

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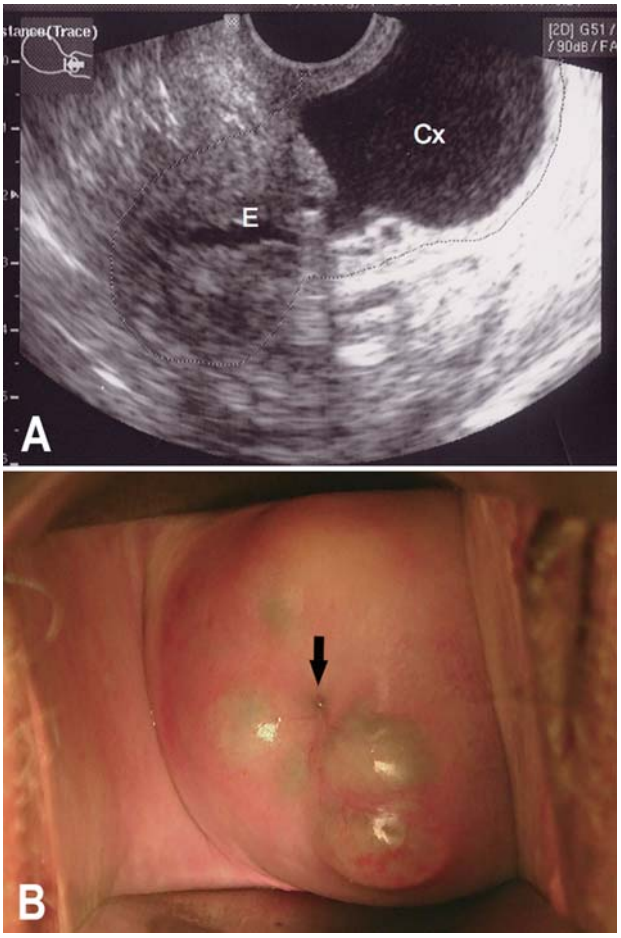

Fig. 1 (**A**) Transvaginal echography from a midline sagittal view demonstrates the markedly distended cervix (Cx) adjacent to the endometrial cavity (E) with a small amount of fluid accumulation in the uterine cavity. (**B**) The speculum examination showed a bulky cervix with complete occlusion of the external ostium (black arrow).

and flat, and no tenderness or palpable mass was found. Transvaginal ultrasound demonstrated a 6-cm well-encapsulated homogeneous hypoechoic cystic mass in the pelvis. After outlining the uterine contour, we uncovered a connection between the uterine cavity and the cyst (Fig. 1A). The right ovary appeared normal with a size of approximately 2.5×1.5 cm and the left ovary was not visualized. Speculum inspection disclosed a bulky cervix with complete occlusion of the external ostium (Fig. 1B), which implied obstruction of the outflow tract and hematotrachelos. Cervical incision of the external ostium with dilation and suction curettage yielded 50 ml of chocolate-like blood clot and confirmed the diagnosis. The end of an Ipas Karman cannula (6 mm diameter and 6 cm length) was inserted into the cervical canal by nylon fixation with simple sutures at 3, 6, 9 and 12 o'clock on the cervix with 1 cm protruding from the external ostium. This was retained for 1 month. Histopathologic findings of the tissue obtained at curettage showed hemorrhage and chronic inflammation. After removing the retained plastic tube 1 month later, the cervical canal was patent. Postoperative recovery was uneventful and the patient has resumed completely regular periods since then.

DISCUSSION

Secondary amenorrhea is the absence of menstrual bleeding for 3 or more months in a woman with previously normal menstruation. The diagnostic process should first exclude physiological states such as pregnancy, lactation (breastfeeding), cycle suppression with systemic contraceptive pills, or menopause. Other differential diagnoses include anatomic defects of the outflow tract, primary hypogonadism (gonadal dysgenesis, gonadal agenesis, or primary ovarian failure), hypothalamic causes (stress, overexercise, or nutrition-related dysfunction), pituitary causes (prolactinoma or autoimmune disease), other endocrine gland disorders (adrenal disease, thyroid disease, or ovarian tumors) and multifactorial causes.[8-10] History, physical examination, and estimation of FSH, thyroid-stimulating hormone (TSH), and prolactin levels will identify the most common causes of amenorrhea. In the present case, the discovery under transvaginal ultrasound of a pelvic cyst with connection to the uterine cavity led to the suspicion of outflow tract obstruction with distended cervix.

Obstruction of the lower female genital tract causes proximal dilatation and development of hematocolpos, hematotrachelos, and hematometra. Symptoms noted in adolescents with congenital obstruction of the lower genital tract usually consist of primary amenorrhea and dysmenorrhea,[1-3] which are not compatible with this patient. In acquired etiologies, the clinical symptoms usually consist of secondary amenorrhea with dysfunctional uterine bleeding, lower abdominal pain, or rarely, acute urine retention.[11,12] The etiologies of acquired obstruction include scarring of the endocervix resulting from iatrogenic trauma such as dilatation and curettage, cone biopsies, loop electrosurgical resection, obstetric laceration, chronic endometriosis, cervical or endometrial carcinoma, or radiation therapy.[13-16] To our knowledge, spontaneously acquired hematotrachelos developing in the perimenopausal period is extremely rare. In this case, the acquired obstruction of lower genital tract was

a result of the patient's nulliparousity and scarring of the endocervix resulting from previous iatrogenic trauma including dilatation and curettage. The perimenopausal effect of cervical atrophy, which is caused by insufficient estrogen, might also play a role in the obstruction of the cervical ostium.

Many Taiwanese women experience perimenopausal amenorrhea that is often left untreated because of the acceptance of menopause as a natural process. There is a general lack of bimanual and speculum examination by nongynecologist physicians, which leads to misdiagnosis of hematotrachelos. Hematotrachelos is often misdiagnosed as an adnexal mass such as ovarian endometrioma or uterine leiomyoma based on ultrasound findings, especially when hematometra is not present. Sherer et al. described hematotrachelos with an hourglass structure demonstrated in a transvaginal ultrasonographic longitudinal scan.[11] The hourglass appearance resulted from simultaneous hematometra and hematotrachelos. However, in the absence of marked distension of the uterus the echographic features in this case differed and had an appearance much more like a lollipop with hematotrachelos alone. As a result, the local clinic diagnosed the patient with ovarian endometrioma without noticing the delicate connection between the uterine cavity and the distended cervix. In this case, combined speculum inspection of the cervix and transvaginal ultrasound helped to identify the hematotrachelos. Placement of a tube into the cervical canal with adequate fixation is necessary to prevent further obstruction of the lower genital tract, particularly for perimenopausal nulliparous women. There is no definitive method for insertion of the tube. Witt introduced the use of a retained latex nasopharyngeal tube as a temporary stent after dilatation and curettage.[17] We tried a new method using a tube for menstrual regulation that is more easily acquired by a gynecological physician. The firm plastic end of an Ipas Karman cannula (6 mm diameter and 6 cm length) with nylon fixation at 3, 6, 9 and 12 o'clock positions and 1 cm protruding from external ostium was retained for 1 month, resulting in a patent cervical canal.

CONCLUSIONS

Hematotrachelos is an uncommon disease and easily misdiagnosed. Detailed history taking, physical examination including speculum examination and recognition of the features on transvaginal ultrasound is the key to diagnosis. A rigid cannula retained as a temporary stent is sufficient treatment.

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