An unusual presentation of usual-type endocervical adenocarcinoma with lobular endocervical glandular hyperplasia: A case report

Yasuteru Sasakura a,⁎, Tetsuya Katsumori a, Osamu Kizu a, Hiroko Yomo b, Masamichi Bamba c

a Department of Radiology, Saiseikai Shiga Hospital, Ohashi 2-4-1, Ritto, Shiga 520-3046, Japan
b Department of Obstetrics and Gynecology, Saiseikai Shiga Hospital, Ohashi 2-4-1, Ritto, Shiga 520-3046, Japan
c Department of Pathology, Saiseikai Shiga Hospital, Ohashi 2-4-1, Ritto, Shiga 520-3046, Japan

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A B S T R A C T

Uterine cervical hematoma is rare. A 51-year-old woman underwent pelvic magnetic resonance imaging (MRI) for uterine tumor survey. A large hematoma with cystic and solid lesions was observed in the uterine cervix. Follow-up MRI after 5 days revealed that the hematoma had decreased in volume. Pathological examination after surgery suggested there was usual-type endocervical adenocarcinoma (UEA) in the lower cervix and lobular endocervical glandular hyperplasia (LEGH) in the upper cervix, along with the cervical hematoma. The findings of this case suggest that the uterine cervical hematoma was secondary to either UEA or LEGH.

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1. Introduction

Uterine cervical hematoma is rare. Causes include: iatrogenic adverse events associated with dilatation and curettage or biopsy [1]; uterine vascular abnormality [2]; uterine cervical tumors, such as endometrial cysts, cervical leiomyoma, and leiomyosarcoma [3,4]; and several pregnancy- or childbirth-related disorders [5,6]. However, a uterine cervical hematoma simultaneously occurring with a usual-type endocervical adenocarcinoma (UEA) or lobular endocervical glandular hyperplasia (LEGH) has never been reported. We report a case of a large cervical hematoma occurring with UEA and LEGH.

2. Case Presentation

A 51-year-old woman (G0P0) presented to hospital with a 4-day history of vaginal bleeding and fever. She had no history of trauma or any medical abnormality in her family. Laboratory examinations showed an elevated serum CA 19–9 level (43.6 U/mL), whereas other tumor markers were normal. On internal examination of the pelvis, it was found that the external ostium of the uterus had stenosis. Transvaginal ultrasonography revealed a 6-cm tumor in the cervix and multiple uterine leiomyomas in the body and fundus. No other abnormalities were found on endometrial cytology or pap smear examination.

Pelvic magnetic resonance imaging (MRI) revealed that the cervical tumor consisted of multiple cysts, which exhibited a so-called cosmos pattern, including a large hematoma predominately located at the upper portion of the cervix. The solid tumor also occupied the lower portion of the cervix (Fig. 1a and b). Although the solid tumor invaded the cervix, it was limited to this area (Fig. 1a and b). Contrast-enhanced MRI 5 days later revealed that the large hematoma had markedly decreased in volume, and the solid tumor was well enhanced (Fig. 1c). Contrast-enhanced computed tomography (CT) scan obtained 14 days later demonstrated that the cervical hematoma had shrunk further (Fig. 1d). On the basis of the imaging findings and clinical information, the cervical tumor was suspected to be gastric-type mucinous carcinoma (GAS). Thereafter, the patient underwent total hysterectomy and bilateral adnexal excision. However, during the operation, surgeons did not find swelling of the uterine cervix suggestive of a tumor.

Macroscopically, the resected specimen revealed multiple cysts in the upper cervix with hemorrhage and a solid tumor in the lower posterior wall of the cervix (Fig. 2a). Microscopically, the cysts in the upper cervix were composed of dilated benign cervical glands with relatively pale cytoplasm, indicating parts of LEGH. The tumor in the lower cervix was composed of invasive growing atypical tubules with pleomorphic nuclei and eosinophilic cytoplasm, indicating UEA (Fig. 2b). One cyst of LEGH had exudates with red blood cells, indicating that it was partially damaged and had been replaced by granulation tissue with hemosiderin
deposits (Fig. 2c). There were no findings suggestive of GAS, sarcoma, or uterine vascular abnormality such as arteriovenous malformation.

Pathological examination revealed that the UEA (Stage I–B1) had invaded the lymph duct. Therefore, additional pelvic lymphadenectomy was performed 11 days after initial surgery. The cervical tumor was finally diagnosed as UEA accompanied by LEGH, with a hematoma occurring simultaneously. The postoperative course was uneventful. Subsequently, the patient underwent chemotherapy and no recurrence was noted over the next 2 years.

3. Discussion

This is a case of a non-traumatic cervical hematoma located in the uterine cervix in a patient with UEA and LEGH. Although the exact mechanism of the formation of hematoma is unclear, pathological examination revealed no other factors causing cervical hematoma, except for UEA or LEGH. Therefore, we considered the lesions as the cause of the hematoma. Bleeding into the vagina or uterine cavity is common in UEA; however, to the best of our knowledge there is no report of a hematoma in the uterine cervix secondary to UEA or LEGH. This is a unique but important finding because it may mimic other uterine cervical tumors such as GAS or sarcoma. It should also be noted that the cervical hematoma rapidly decreased in volume. However, we could not determine the exact mechanism of this finding.

UEA is the second most common type of cervical cancer and accounts for 25% of all cervical cancers [7]. The majority of cervical adenocarcinoma is UEA, which is composed of solid lesions. The MRI features of UEA have been reported as being an invasive solid or diffuse infiltrative pattern [4]. Depending on the stage, UEA is treated by surgery, radiation therapy, or chemotherapy [8].

LEGH is a benign lesion that develops in women at a mean age of 45–48 years [9,10], occasionally with increased watery discharge and
Fig. 2. (a) Macroscopic view of the resected specimen. Multiple cysts in the upper cervix with hemorrhage (arrows) and a solid tumor in the lower posterior wall of the cervix (arrowheads) were noted. The stars indicate the uterine leiomyoma. (b) Hematoxylin eosin and staining (x5 magnification) of the specimen obtained after hysterectomy. The cysts in the upper cervix were composed of dilated benign cervical glands with relatively pale cytoplasm (arrows), suggesting parts of lobular endocervical glandular hyperplasia (LEGH), whereas the solid tumor that extended near LEGH was composed of invasive growing atypical tubules with pleomorphic nuclei and eosinophilic cytoplasm (arrowheads), suggesting usual-type endocervical adenocarcinoma (UEA). (c) Hematoxylin eosin and staining (x20 magnification) of the specimen obtained after hysterectomy. The LEGH walls were partially damaged and had formed granulation tissue with hemosiderin deposits (arrow), but there were no invasive atypical tubules suggestive of UEA in the walls. The LEGH included exudates with red blood cells (arrowheads). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)
cervical mass. On MRI, LEGH shows small cysts surrounded by large cysts (the so-called cosmos pattern) near the internal ostium of the uterus [11,12]. LEGH is considered as a precursor lesion to GAS [13,14]. Although it is difficult to differentiate between GAS and LEGH, the presence of a solid lesion in the cysts is indicative of GAS [11,12,15]. Therefore, careful follow-up or surgery is performed, depending on the results of MRI and pap smear [11,15].

In summary, the present case demonstrated that the uterine cervical hematoma was most likely induced by either UEA or LEGH, and can be added as differential diagnosis in cervical hematoma. The mechanisms of hematoma formation and the abrupt decrease in volume remain unclear. Further studies are necessary to understand these processes.

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