Case Reports in Women's Health 30 (2021) e00297

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

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

ARTICLE INFO

Article history:
Received 15 January 2021
Received in revised form 28 January 2021
Accepted 2 February 2021
Available online xxxx

Keywords:
Cervical hematoma
Usual-type endocervical adenocarcinoma
Uterus
Lobular endocervical glandular hyperplasia
Magnetic resonance imaging

ABSTRACT

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.

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

* Corresponding author.
E-mail address: sasakura@koto.kpu-m.ac.jp (Y. Sasakura).

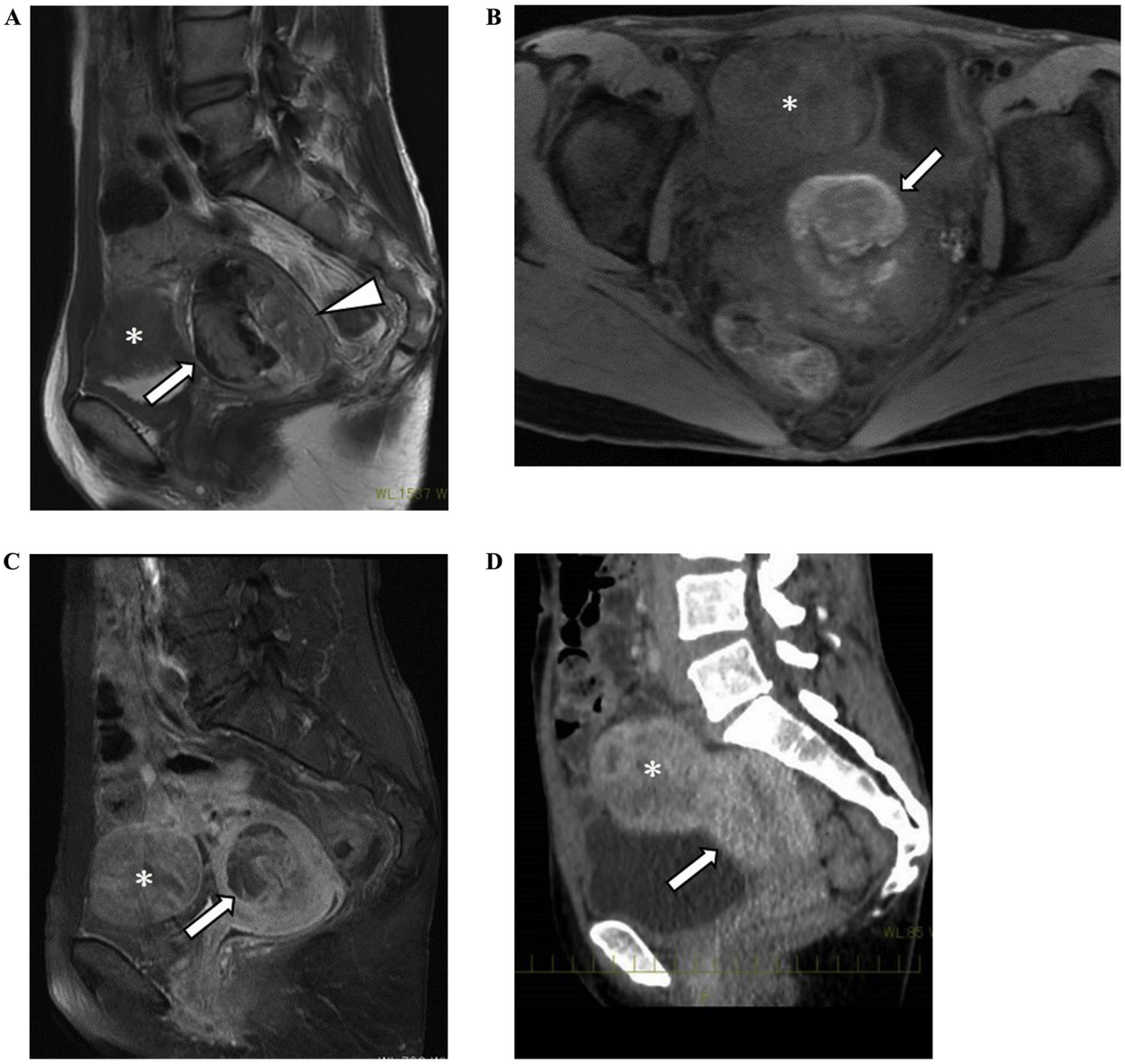

Fig. 1. (a) Sagittal T2-weighted image at baseline. (b) Axial fat-saturated T1-weighted image at baseline. The cervical tumor was composed of multiple cysts, including a large hematoma (arrow) predominately located at the upper portion of the cervix (a and b) and the solid tumor (arrowhead) located at the lower portion (a). The cysts in the upper cervix included bloody fluid collection/hematoma, which was observed as low signal on the sagittal T2-weighted image (a) and as high signal on the axial T1-weighted image (b). The solid tumor in the lower cervix was limited to the cervix. The star indicates the uterine leiomyoma. (c) Sagittal contrast-enhanced fat-saturated T1-weighted image 5 days later. The hematoma in the cervix markedly decreased in volume. The hematoma exhibited low signal. The star indicates the uterine leiomyoma. (d) Sagittal contrast-enhanced CT 14 days later. The hematoma in the cervix markedly decreased in volume. The star indicates the uterine leiomyoma.

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

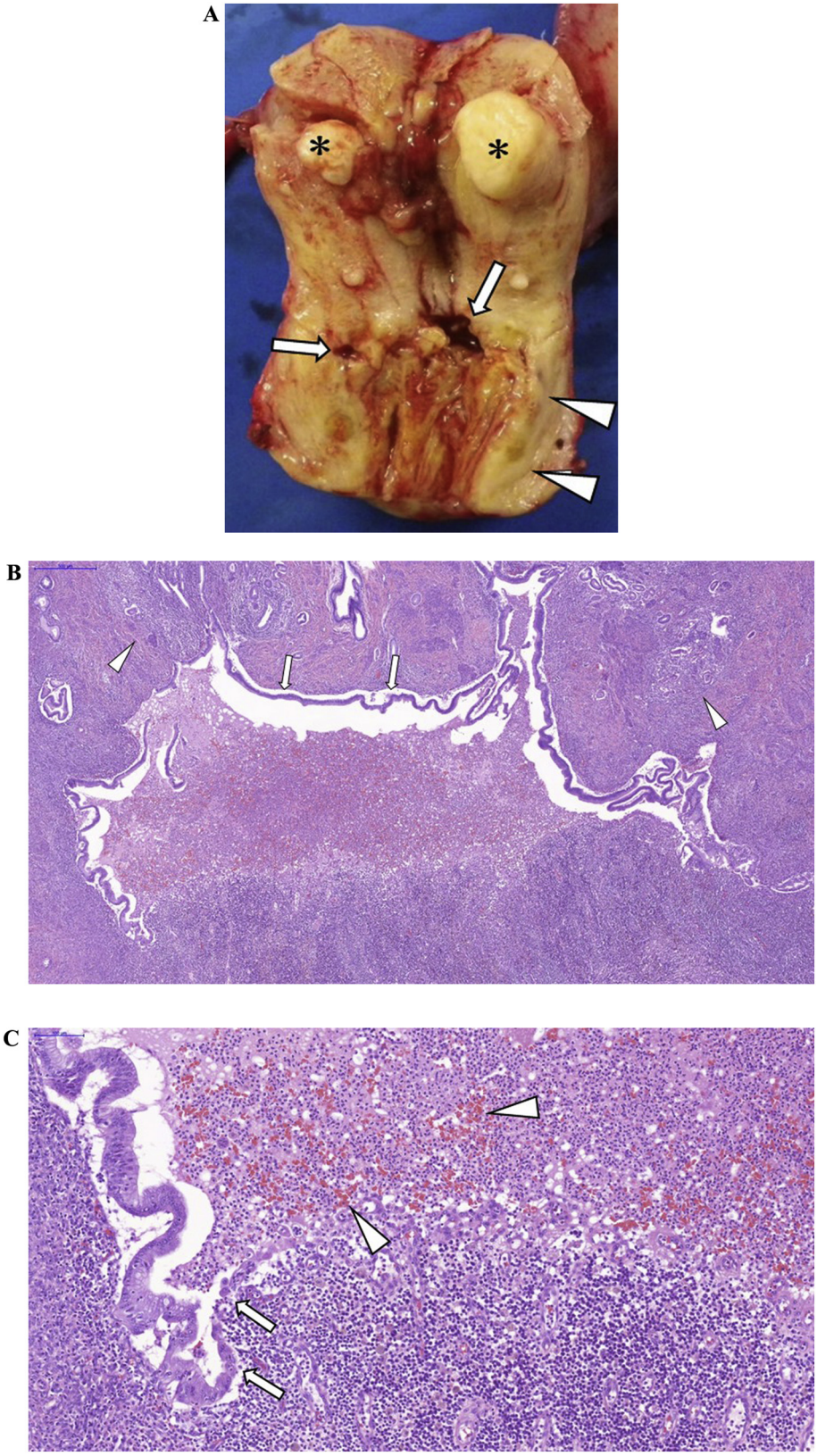

Fig. 2. (a) Macroscopic view of the resected specimen. Multiple cysts in the upper cervix with hemorrhage (arrows) and a solid tumor in the lower posterior wall of the cervix (arrowheads) were noted. The stars indicate the uterine leiomyoma. (b) Hematoxylin eosin and staining (x5 magnification) of the 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.

Contributors

Yasuteru Sasakura contributed to data curation, resources, and writing the original draft.

Tetsuya Katsumori contributed to drafting the manuscript, review, editing and supervision.

Osamu Kizu contributed to drafting the manuscript, review and editing.

Hiroko Yomo was responsible for conceptualization and acquisition of data.

Masamichi Bamba reviewed pathology and prepared the manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

No funding from an external source supported the publication of this case report.

Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

Acknowledgments

We would like to thank Dr. Tatsuya Yoshikawa, Department of Radiology, Saiseikai Shiga Hospital, for help with preparation of this manuscript.

References

[1] H.S. Wong, Y.K. Cheung, K. Burns, J. Hawley, A case of uterine hematoma presenting as a transient vascular abnormality on ultrasonography, Ultrasound Obstet. Gynecol. 26 (2005) 682–683, https://doi.org/10.1002/uog.2603.

[2] S. Soeda, J. Ushijima, S. Furukawa, M. Miyajima, K. Sakuma, T. Watanabe, M. Miyazaki, Y. Hashimoto, H. Nishiyama, K. Fujimori, Uterine arteriovenous malformation formed in a large uterine cervical myoma, Tohoku J. Exp. Med. 228 (2012) 181–187, https://doi.org/10.1620/tjem.228.181.

[3] N. Yokota, H. Yoshida, H. Sakakibara, Y. Inayama, F. Hirahara, A severe vaginal hemorrhage caused by cervical endometriosis, Am. J. Obstet. Gynecol. 199 (2008) 12–13, https://doi.org/10.1016/j.ajog.2008.02.012.

[4] T. Saida, A. Sakata, Y.O. Tanaka, H. Ochi, T. Ishiguro, M. Sakai, H. Takahashi, T. Satoh, M. Minami, Clinical and MRI characteristics of uterine cervical adenocarcinoma: its variants and mimics, Korean J. Radiol. 20 (2019) 364–377, https://doi.org/10.3348/kjr.2018.0458.

[5] Y. Okamoto, Y.O. Tanaka, M. Nishida, H. Tsunoda, H. Yoshikawa, Y. Itai, MR imaging of the uterine cervix: imaging-pathologic correlation, Radiographics 23 (2003) 425–445, https://doi.org/10.1148/rg.232025065.

[6] S.E. Jung, J.Y. Byun, J.M. Lee, B.G. Choi, S.T. Hahn, Characteristic MR findings of cervical pregnancy, J. Magn. Reson. Imaging 13 (2001) 918–922, https://doi.org/10.1002/jmri.1131.

[7] H.O. Smith, M.F. Tiffany, C.R. Qualls, C.R. Key, The rising incidence of adenocarcinoma relative to squamous cell carcinoma of the uterine cervix in the United States - a 24-year population-based study, Gynecol. Oncol. 78 (2000) 97–105, https://doi.org/10.1006/gyno.2000.5826.

[8] NCCN Clinical Practice guidelines in Oncology, Pancreatic Adenocarcinoma Version 4, https://www.nccn.org/professionals/physician_gls/pdf/cervical.pdf 2019 (accessed 21 January 2021).

[9] M.R. Nucci, P.B. Clement, R.H. Young, Lobular endocervical glandular hyperplasia, not otherwise specified: a clinicopathologic analysis of thirteen cases of a distinctive pseudoneoplastic lesion and comparison with fourteen cases of adenoma malignum, Am. J. Surg. Pathol. 23 (1999) 886–891, https://doi.org/10.1097/00000478-199908000-00005.

[10] Y. Mikami, S. Hata, J. Melamed, K. Fujiwara, T. Manabe, Lobular endocervical glandular hyperplasia is a metaplastic process with a pyloric gland phenotype, Histopathology. 39 (2001) 364–372, https://doi.org/10.1046/j.1365-2559.2001.01239.x.

[11] A. Takatsu, T. Shiozawa, T. Miyamoto, K. Kurosawa, H. Kashima, T. Yamada, T. Kaku, Y. Mikami, T. Kiyokawa, H. Tsuda, K. Ishii, K. Togashi, T. Koyama, Y. Fujinaga, M. Kadoya, A. Hashi, N. Susumu, I. Konishi, Preoperative differential diagnosis of minimal deviation adenocarcinoma and lobular endocervical glandular hyperplasia of the uterine cervix: a multicenter study of clinicopathology and magnetic resonance imaging findings, Int. J. Gynecol. Cancer 21 (2011) 1287–1296, https://doi.org/10.1097/IGC.0b013e31821f746c.

[12] A. Ohya, S. Asaka, Y. Fujinaga, M. Kadoya, Uterine cervical adenocarcinoma associated with lobular endocervical glandular hyperplasia: radiologic–pathologic correlation, J. Obstet. Gynaecol. Res. 44 (2018) 312–322, https://doi.org/10.1111/jog.13528.

[13] Y. Mikami, T. Kiyokawa, S. Hata, K. Fujiwara, T. Moriya, H. Sasano, T. Manabe, J.I. Akahira, K. Ito, T. Tase, N. Yaegashi, I. Sato, H. Tateno, H. Naganuma, Gastrointestinal immunophenotype in adenocarcinomas of the uterine cervix and related glandular lesions: a possible link between lobular endocervical glandular hyperplasia/pyloric gland metaplasia and "adenoma malignum", Mod. Pathol. 17 (2004) 962–972, https://doi.org/10.1038/modpathol.3800148.

[14] A. Takatsu, T. Miyamoto, C. Fuseya, A. Suzuki, H. Kashima, A. Horiuchi, K. Ishii, T. Shiozawa, Clonality analysis suggests that STK11 gene mutations are involved in progression of lobular endocervical glandular hyperplasia (LEGH) to minimal deviation adenocarcinoma (MDA), Virchows Arch. 462 (2013) 645–651, https://doi.org/10.1007/s00428-013-1417-1.

[15] S. Bin Park, J.H. Lee, Y.H. Lee, M.J. Song, K.T. Lim, S.R. Hong, J.K. Kim, Adenoma malignum of the uterine cervix: imaging features with clinicopathologic correlation, Acta Radiol. 54 (2013) 113–120, https://doi.org/10.1258/ar.2012.120059.