



Case Report

Complete non-puerperal uterine inversion caused by uterine hemangioma: A case report

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ABSTRACT

Objective: Uterine inversion is a rare postpartum complication. Non-puerperal uterine inversion is extremely rare. It mostly occurs with uterine tumors, especially leiomyoma. In most instances, the inversion may not be noticed until the time of surgery. The preoperative diagnosis is difficult.

Case report: We report a case of non-puerperal complete uterine inversion that was initially diagnosed as cervical cancer. The uterine inversion was diagnosed preoperatively and she underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy. The histological examination showed uterine hemangioma.

Conclusion: Accurate diagnosis of the non-puerperal uterine inversion is important. Surgical intervention is necessary and it provides good prognosis. Hemangioma may be one of the causes of non-puerperal uterine inversion.

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Introduction

Uterine inversion is a rare clinical problem. Puerperal uterine inversion occurs with an incidence of 1 in 3500 deliveries and is encountered as an obstetric emergency [1]. In non-puerperal instance, uterine inversion is extremely rare. Most cases of non-puerperal uterine inversion may not be noticed until the time of surgery. It still remains a diagnostic challenge in gynecology. The vast majority of non-puerperal inversion cases are associated with tumors of the corpus uterus. Fibroid is the most common cause, carcinoma and sarcoma were also being reported [2]. In the literature, uterine inversion secondary to uterine hemangioma has never been reported before.

Here we report our experience in the diagnosis and treatment of the first case of complete non-puerperal uterine inversion associated with uterine hemangioma.

Case presentation

A 44-year-old woman, gravida 2, para 2, suffered from intermittent vaginal bleeding for two months. She visited a local clinic due to acute abdominal cramping pain and was then referred to our hospital with the diagnosis of bulky cervical cancer. On speculum examination, a large mass lesion occupying the vaginal cavity was observed and the normal uterine cervix contour could not be visualized. The mass showed necrotic appearance with contact bleeding. Foul smelling purulent discharge was also present. On bimanual examination, severe uterine tenderness was noted and the size of the uterus was slightly enlarged with irregular shaped fundus. The patient was anemia (Hb 5.1 g/dL). Leukocytosis (WBC 17100/cumm) was also noted. For the nature of the vaginal mass, direct punch biopsies were performed. But they failed to show any malignancy, only necrotic tissue and spindle cells of uncertain origin found. On transabdominal ultrasonographic examination, the vaginal mass was assessed as 13 cm × 8.5 cm × 8.7 cm in size. The shape of endometrium could not be traced. But the longitudinal scan showed an indentation and a longitudinal hypoechoic groove at

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the uterine fundus (Fig. 1). A large delivery myoma with uterine inversion was suspected and operation intervention was decided.

At surgery, central dimpling of the fundal uterus was found. Fallopian tubes and ovaries protruded from the edge of the dimpling, and they are congestive. Complete uterine inversion was proved (Fig. 2). Initially we attempted the Huntington operation to revert the uterus, but the vaginal mass was so large that could not pass the cervical os and the uterus could not be restored to its normal shape. We amputated the mass transvaginally and soon performed total abdominal hysterectomy and bilateral salpingo-oophorectomy (Fig. 3) smoothly with blood loss about 350 ml. There was no other intraoperative complication noted. The specimen showed a protruding tumor measuring 10 cm × 8 cm × 6.5 cm over the inverted uterine fundus. Its outer surface is necrotic, and on cut it showed areas of hemorrhage. Grossly there was no definite myoma found. In the histological examination, the myometrium at the uterine fundus showed irregular distribution of varying-calibered vascular channels containing organizing thrombi (Fig. 4). Cavernous hemangioma was diagnosed. The patient tolerated the surgery well and was discharged 5 days later.

Discussion

Uterine inversion is uncommon; it is more often encountered as an obstetric emergency due to postpartum hemorrhage. In non-obstetric instances, uterine inversion is extremely rare, with only 150 cases reported from 1887 to 2004 in a literature review [3]. According to the degree of inversion, it can be classified as incomplete, complete, or total. Das reported 47 cases of tumor-related non-puerperal uterine inversion, of which 87% were due to fibroids, 7.4% to sarcoma, and 5.6% to carcinoma [2]. Uterine hemangioma is rare, with fewer than 50 cases reported in literature. Large hemangiomas can extend through the full thickness of the myometrium and result in severe bleeding. In the literature, uterine inversion secondary to uterine hemangioma has never been reported.

Possible mechanisms of non-puerperal uterine inversion were proposed [2]. Most people agree with thinning and weakening of



Fig. 1. The longitudinal ultrasound scan showed an indentation (white arrow) and a longitudinal hypoechoic groove (black arrow) at the uterine fundus, which provided the diagnosis of uterine inversion. The arrowheads outline the protruding mass.



Fig. 2. Inversion of the fundal uterus with Fallopian tubes and ovaries protruding from the dimpling.

the uterine wall at the tumor's implantation along with concurrent contractions of the uterine musculature expelling the tumor through the cervix into the vagina. Weight and size of the tumor, cough and sneezing put traction on the thinned uterine wall may also aggravate the inversion [4]. From our case, we think that hemangioma may also soften the uterine fundus and cause the uterine inversion.

Most cases of non-puerperal uterine inversion are chronic. The presenting symptoms are vaginal bleeding and discharge, pelvic pain, mass protruding, and urinary disturbance. Our patient suffered from most of the above symptoms for about two months. Gynecologic malignancy should be precluded before operation. In Taiwan, bulky advanced cervical cancer is still occasionally seen,

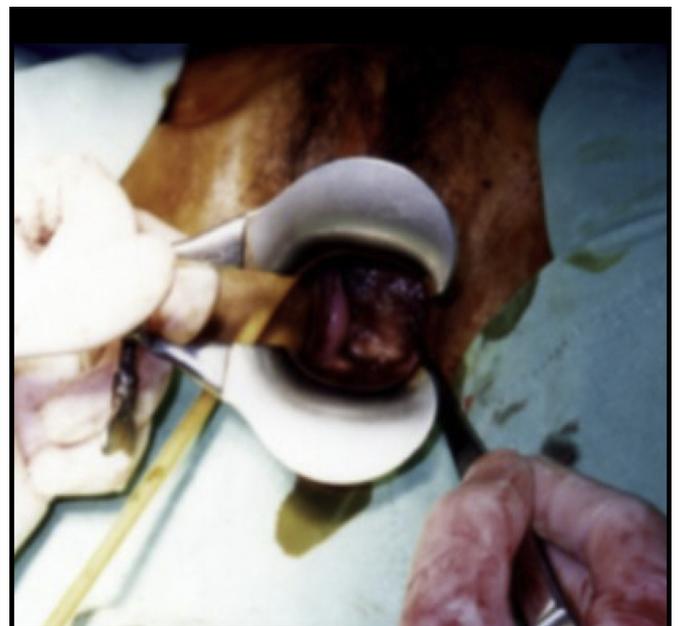


Fig. 3. The protruding mass can be accessed by speculum exam. The hemangioma was amputated from vagina and ATH + BSO was performed subsequently.

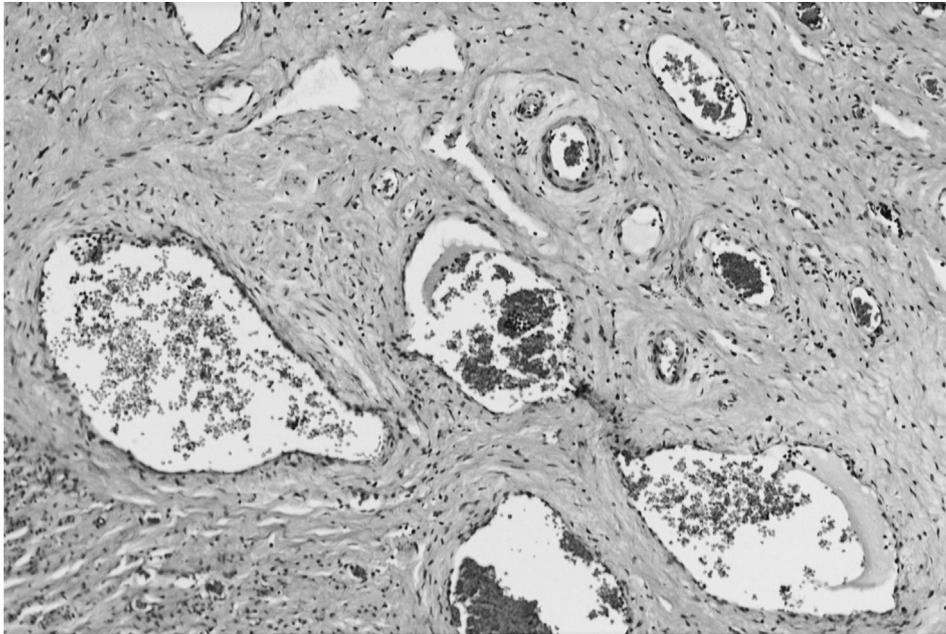


Fig. 4. Histological finding of the fundal myometrium showed irregular distribution of varying-calibered vascular channels containing organizing thrombi. Hematoxylin and eosin (H&E), $\times 400$.

especially in those patients who never received cervical smear or any other gynecological examinations before. Our patient searched for help late after suffering from massive bleeding, abdominal cramping, and severe anemia. Her clinical appearance was quite similar to that of advanced cervical cancer. Nevertheless, cervical cancer was precluded after biopsy of the vaginal mass.

Clinical diagnosis of uterine inversion is difficult unless the fundal depression can be palpated in bimanual examination, and its presence may not be noticed until the time of surgery [5]. In postpartum partial uterine inversion, a Y-shaped uterine cavity in the longitudinal plane and a “bull-eye” appearance of the transverse view of the corpus can be detected in ultrasound examination due to the infolding of the endometrial surface [6]. In non-puerperal uterine inversion, such typical appearance may be obscured by distorted uterine anatomy and the uterine tumor. An indentation with a longitudinal hypoechoic groove of the fundal uterus under transabdominal ultrasound on a longitudinal scan was also reported as the suspicious finding of inversion [7]. In our case, this finding provided us with the preoperative diagnosis of uterine inversion. Magnetic resonance imaging (MRI) is useful not only in diagnosis but also to delineate the lesion in neighbor structures [8]. Lewin et al. reported that a U-shaped uterine cavity and a thickened and inverted fundus on a T2-weighted image in a sagittal view are indicative of uterine inversion [9]. Preoperative diagnosis of hemangioma is difficult. Vaginal examination, uterine curettage specimens, ultrasonography, and hystero-graphy are usually uninformative. Pelvic angiography or computed tomography could confirm the vascular nature of the lesion. An ultrasound-guided biopsy could be helpful.

Treatment of non-puerperal uterine inversion depends on the preoperative diagnosis. Because the possibility of uterine malignancy, and the large majority of post-reproductive age, hysterectomy is the treatment of almost all reported cases. The Huntington operation is the first choice before hysterectomy [10]. It involves grasping the round ligaments and uterus below the area of inversion and slowly pulling upward repeatedly to revert the uterus. In non-puerperal uterine inversion, some authors suggest transvaginal excision of the tumor mass before

hysterectomy [2]. Combined laparoscopic and vaginal approach was also introduced [11]. In our case, the inverted uterine fundus is so big and fragile that we failed to restore its normal anatomy. Hence, we decided to amputate the inverted uterine fundus transvaginally, which made the following separation of the urinary bladder and ligations of the uterine vasculature much easier. Then, we completed the hysterectomy and bilateral salpingo-oophorectomy without any complication.

Summary

Non-puerperal uterine inversion is extremely rare; it should be taken into consideration in the differential diagnosis in the evaluation of patients with pelvic mass, especially those with protruding mass in the vagina. It is important to make the accurate preoperative diagnosis in order to provide appropriate treatment. This case also suggested that uterine hemangioma might be one of the causes associated non-puerperal uterine inversion.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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References

- [1] Abouleski E, Ali V, Joumaa B, Lopez M, Gupta D. Anaesthetic management of acute puerperal inversion. *Br J Anaesth* 1995;75(4):486–7.
- [2] Gowri V. Uterine inversion and corpus malignancies: a historical review. *Obstet Gynecol Surv* 2000;55(11):703–7.
- [3] Chou WY, Chang HW. Uterine hemangioma: a rare pathologic entity. *Arch Pathol Lab Med* 2012;136(5):567–71.
- [4] Lupovitch A, England ER, Chen R. Non-puerperal uterine inversion in associated with uterine sarcoma: case report in a 26-year-old and review of the literature. *Gynecol Oncol* 2005;97(3):938–41.

- [5] Moodley M, Moodley J. Non-puerperal uterine inversion associated with uterine sarcoma: clinical management. *Int J Gynecol Cancer* 2003;13(2):244–5.
- [6] Gross RC, McGahan JP. Sonographic detection of partial uterine inversion. *AJR Am J Roentgenol* 1985;144:761–2.
- [7] Hsieh TT, Lee JD. Sonographic findings in acute puerperal uterine inversion. *J Clin Ultrasound* 1991;19:306–9.
- [8] Auber M, Darwish B, Lefebure A, Ness J, Roman H. Magnetic resonance imaging of non-puerperal complete uterine. *Iran J Radiol* 2015;12(4):e9878.
- [9] Lewin JS, Bryan PJ. MR imaging of uterine inversion. *J Comput Assist Tomogr* 1989;13(2):357–9.
- [10] Lascarides E, Cohen M. Surgical management of non-puerperal inversion of the uterus. *Obstet Gynecol* 1968;32(3):376–81.
- [11] Auber M, Darwish B, Lefebure A, Ness J, Roman H. Management of non-puerperal uterine inversion using a combined laparoscopic and vaginal approach. *Am J Obstet Gynecol* 2011;204(6):e7–9.