## What is your diagnosis?

A 40-year-old lady with previous two deliveries by lower segment caesarian section presented with a long history of lower abdominal heaviness and menorrhagia and a mass coming out of vagina recently. There were associated problems in voiding urine. On local examination, a large, elongated, soft, reddish mass, measuring  $14 \times 3.8 \times 4$  cm, was protruding out of the vagina. A firm whitish mass with a well-demarcated rim was observed at the end of the large reddish mass, measuring  $5 \times 5$  cm. The entire mass could be incompletely reduced inside the vagina. On genital examination, the cervical rim could not be posteriorly felt. There was ulceration with discharge of pus from the top of the mass, which was sent for culture and sensitivity tests. The culture was positive for Pseudomonas aeruginosa, which was sensitive to levofloxacin. Prolapsed part was reposited back and was restrained using a glycerine acriflavine-soaked vaginal tampon. The patient was put on oral antibiotics with a dressing of the local wound. Healing was achieved in 10 days. An ultrasonography of the pelvis was ordered. Ultrasonography revealed an upside-down uterine fundus, filling in between the cervix making mirror image of normal uterine contour. Outer serosal surfaces were coming in contact together making typical pseudo stripe sign. The uterine fundus was observed between the vaginal walls giving a typical target sign. There was a heterogeneous mass lesion observed at the uterine fundus that probably arised from the submucosa and exhibited heterogeneous internal vascularity. The uterine artery pedicle and ovaries of both the sides were pulled along with the uterine fundus; however, there was no involvement of the urinary bladder (Figure 1 a-d).



Figure 1. a-d. Pelvic ultrasound image (coronal view) shows target sign (solid arrow), i.e., hyperechoic fundus with mass surrounded by hypoechoic layer of fluid inside the vagina (a), pelvic color mode ultrasound image (sagittal view) shows inverted fundus with pulled pedicles (solid arrow) (b), pelvic ultrasound image (sagittal view) shows pseudo stripe sign (solid arrow), i.e., outer serosal surfaces coming into contact mimicking normal endometrial stripe (c), pelvic spectral Doppler mode ultrasound image (sagittal view) shows a normal uterine artery waveform in pulled uterine vascular pedicle (d)

## Answer

The chronic non-puerperal uterine inversion is an extremely uncommon entity, generally caused by submucosal fibroid, particularly located in the fundus (1). Chronic non-puerperal uterine inversion is generally observed after the age of 40 years, and in addition to large submucous fibroids, rare malignancies, such as rhabdomyosarcoma, endometrial carcinoma, or endometrial polyp, can precede etiologic agents (2). The proposed pathogenesis is thinned out uterine wall in middle-aged patients, giant fundal mass, and gravity with expulsive efforts by uterus (3). By definition, uterine inversion means a descent of the uterine fundus to or through the cervix so that the uterine cavity is anatomically turned inside out (4).

Uterine inversion can be divided into acute and chronic on the basis of the onset and course of the disease. Acute inversion is characterized by pain and hemorrhage, whereas chronic inversion is associated with pelvic discomfort, vaginal discharge, irregular vaginal bleeding, and anemia (5).

Uterine inversion is suspected when gynecological examination detects a protruding mass in the vagina or vulva, and the uterine fundus cannot be palpated by bimanual examination. A constricting ring around the cervix is a convincing clinical finding. The most promising features are inability to palpate the fundus and non-visualization of the cervix separately (5). Acute inversion of the uterus is almost always postpartum and is common, whereas chronic inversion of the uterus is extremely rare. Chronic uterine inversion can be either incomplete or complete. In incomplete type, the fundus everts out of the cervix but stays inside the vagina, whereas in complete type, the fundus, including the cervix, protrudes out of the introitus (6). A high index of suspicion is required as chronic uterine inversion can be mistaken for uterine prolapse or any mass of the vagina or cervix. Correct diagnosis can only be made intra-operatively in many cases. This causes certain surgical difficulties and increased complication rates (7).

Ultrasonography generally is the primary diagnostic modality of choice (7). Classical findings described are the mirror image sign, pseudo stripe sign, and target sign. All these features are present in our case. Mirror-image sign depicts the reverse relationship of the fundus and cervix as the fundus lies lower than the cervix. The pseudo stripe sign is described as a hyperechoic stripe formed because of the apposition of serosal surfaces mimicking the normal endometrial stripe. The target sign is an illustration of the uterine fundus lying in the vagina and outlined by hypoechoic line of fluid collection between the two (8).

Characteristic magnetic resonance imaging (MRI) findings are U-shaped alignment of the uterine cavity in sagittal plane and bull's eye appearance of the fundus inside the vagina on T2-weighted imaging, which is a derivative of the target sign found on ultrasonography. MRI also shows the content of the inversion, such as the ovaries or bladder. MRI is the best modality for the characterization of the mass that is responsible for the inversion, i.e., to differentiate between benign and malignant masses (9).

In this case, MRI revealed an U-shaped configuration of the uterine cavity with complete inversion of the fundus. The uterine artery pedicle and ovaries were also pulled along with



Figure 2. a-d. Axial T2-weighted MRI shows Bull's eye sign (solid arrow), i.e., hypointense fundus outlined by hyperintense layer of fluid within the vagina (a), sagittal T2-weighted MRI shows inverted U-shaped uterine cavity (solid arrow), a mirror image of a normal anatomical configuration (b), coronal T2-weighted MRI shows inverted uterus with a well-defined mass lesion at the top of the uterine fundus (solid arrow) (c), sagittal post contrast T1-weighted MRI shows intense homogeneous enhancement of both the mass and normal uterus (d)



Figure 3. a-d. Clinical pre-operative photograph of the patient, showing exposed red fleshy endometrium with a whitish fleshy mass (solid arrow) at top of it (a), intra-operative photograph shows undergoing excision of the fibroid and vaginal myomectomy (b), intra-operative photograph shows undergoing Kustner's procedure, incision at the posterior cervical lip, and identifying edges of the cervix (c), intra-operative photograph shows repositioning of the uterus for vaginal hysterectomy (d)

the uterine fundus. There was a well-defined heterogeneous mass at the uterine fundus revealing a heterogeneous intense enhancement that was suggestive of a fibroid. The U-shaped configuration was previously described in the sagittal plane and can be observed in the coronal plane, similar to this case (Figure 2 a-d).

Huntington and Haultain procedures are the commonly used abdominal approaches, and Kustner and Spinelli procedures are the commonly used vaginal approaches for the correction of chronic inversion (10).

As the fibroid was present at the fundus; therefore, vaginal myomectomy was first performed and then the uterus was reposited so that safe hysterectomy could be performed. Kustner's procedure was performed as the patient had history of two deliveries by lower segment caesarian section, and there are less chances of injury to the bowel and bladder in this procedure (Figure 3 a-d). The patient had a normal course in the ward and was discharged after 5 days.

The following conclusions can be drawn from this case. This is a very rare entity with typical imaging features; however, many radiologists are unaware of this entity because of its rarity. This differential it must be kept in mind in middle-aged patients presenting with a mass coming out of the vagina. As the method of treatment solely depends on the perfect radiological diagnosis, precise diagnosis must be delivered keeping in mind the abovementioned imaging features.

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## References

- De Vries M, Perquin DA. Non-puerperal uterine inversion due to submucous myoma in a young woman: a case report. J Med Case Rep 2010; 4: 21. [CrossRef]
- Lupovitch A, England E.R, Chen R. Non-puerperal uterine inversion in association with uterine sarcoma: case report in a 26-year-old and review of the literature. Gynecol Oncol 2005; 97: 938-41. [CrossRef]
- Krenning RA, Dörr PJ, de Groot WH, de Goey WB. Non-puerperal uterine inversion. Case report. Br J Obstet Gynaecol 1982; 89: 247-9.
  [CrossRef]
- 4. Lai FM, Tseng P, Yeo SH, Tsakok FH. Non puerperal uterine inversion: a case report. Singapore Med J 1993; 34: 466-8.
- Kopal S, Seckin N.C, Turhan N.O. Acute uterine inversion due to a growing submucous myoma in an elderly woman: case report. Eur J Obstet Gynecol Reprod Biol 2001; 99: 118-20. [CrossRef]
- Jain S, Aherwar R, Joshi P. Chronic Non-Puerperal Uterine Inversion; Fibromyoma Uterias a Cause- A Case Report. Sch J Med Case Rep 2014; 2: 100-2.
- Hu C.F, Lin H. Ultrasound diagnosis of complete uterine inversion in a nulliparous woman. Acta Obstet Gynecol Scand 2012; 91: 379-81. [CrossRef]
- 8. Rana KA, Patel PS. Complete Uterine Inversion An Unusual Yet Crucial Sonographic Diagnosis. J Ultrasound Med 2009; 28: 1719-22.
- Occhionero M, Restaino G, Ciuffreda M, Carbone A, Sallustio G, Ferrandina G. Uterine inversion in association with uterine sarcoma: a case report with MRI findings and review of the literature. Gynecol Obstet Invest 2012; 73: 260-4. [CrossRef]
- 10. Lascarides E, Cohen M. Surgical management of the nonpuerperal inversion of the uterus. Obstet Gynecol 1968; 32: 376-81.