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CASE REPORT

Non-Puerperal Uterine Inversion: A Case Report

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ABSTRACT

An extremely rare case of complete non- puerperal uterine inversion caused by a low grade endometrial stromal sarcoma at the fundus in a 45-year-old divorced woman presenting with profuse vaginal bleeding was reported. The preoperative diagnosis was performed by using full radiological investigations,i.e. computed tomography(CT), ultrasonography, and magnetic resonance imaging (MRI). Total abdominal hysterectomy with bilateral salpingo-oophorectomy was carried out following successful reversion of the uterine inversion by Haultian procedure.

Key words: Uterine inversion, non-puerperal, Haultain procedure

Uterine inversion is prolapse of the fundus to or through the cervix so that the uterus is in effect turned inside out.[1] In 1951, Jones classified uterine inversion into two types, the first one is puerperal or obstetric and the other one is non-puerperal or gynecologic.[2]

Non-puerperal or gynecologic uterine inversion is extremely rare.[3] Most of these condition are associated with either benign or malignant tumor arising from the uterine fundus such as submucous leiomyoma, sarcoma and endometrial carcinoma[4] and is considered idiopathic when there is no recognizable cause. [2,5]

Cases of complete and total uterine inversion are usually accompanied by constriction of the external cervical os that produce vascular stasis and in neglected cases, gangrene of the inverted uterus will be occurred.[4]

We present the first case of complete non-puerperal uterine inversion in the literature review that was diagnosed preoperatively with full investigation of imaging, i.e. computerized tomographic-scan (CT-scan), ultrasonography, and magnetic resonance imaging (MRI).

Case Report

The patient was a 45-year-old divorced woman, para 1011,last abortion 11 years ago. The first gravida

resulted in normal labor with male fetus17 years ago. The second one was spontaneous abortion at 2 months of gestation and the patient had undergone dilatation and curettage with no complication. After that she never had seen the doctor.

The patient had abnormal vaginal discharge for 3 months prior to admission on July 28, 1998. Three days before admission, she had profuse vaginal bleeding and lower abdominal pain after riding a motorcycle. Physical examination revealed marked anemia. Pelvic examination showed an exophytic mass with necrotic tissue about eight cms. in diameter, seemed to involve the whole cervix but cervical os could not be identified. The uterus was not palpable in the pelvis,both adnexa and parametria were normal. Cervical punch biopsy was done with the provisional diagnosis of cervical cancer stage IB2 according to FIGO staging system. The basic laboratory investigations appeared normal except the hemoglobin was only four gm%. She was subjected to undergo CT-scanning to evaluate the retroperitoneal lymph nodes but the result showed uterine inversion (Fig.1) with no enlargement of any retroperitoneal lymph nodes. However, to confirm and study this rare condition, ultrasonography (Fig.2A) and MRI (Fig.2B) were performed, both of them, especially the MRI showed typical image of uterine inversion with a mass on top.

The pathology of punch biopsy was firstly misinterpreted as leiomyoma. Blood transfusion was administered to correct anemia and antibiotic was given to combat infection. The patient had undergone total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH&BSO) on August 20, 1998 which showed completed uterine inversion with round ligaments and some part of the fallopian tubes embedded in the concavity of the inversion (Fig.3). Huntington procedure was used to reposition the inverted uterus, but failed, so we proceed to the Haultain procedure by performing longitudinal incision along the lower posterior uterine wall (Fig 4). The uterine fundus could be successfully reverted. On gross examination, an exophytic mass, four cms. in diameter with necrotic surface was found at the fundus. The other organs were normal including retroperitoneal lymph nodes. Post operative course was uneventful. The final pathological report low grade stromal sarcoma with focal smooth muscle differentiation and superficial myometrial invasion. She was scheduled to attend regular follow up at gynecologic-oncology clinic.

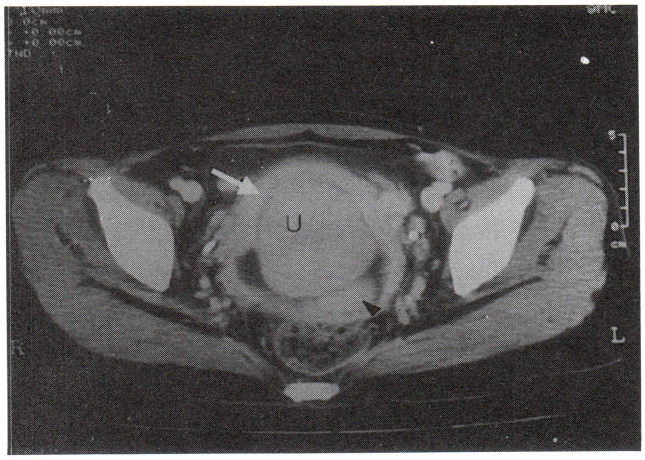
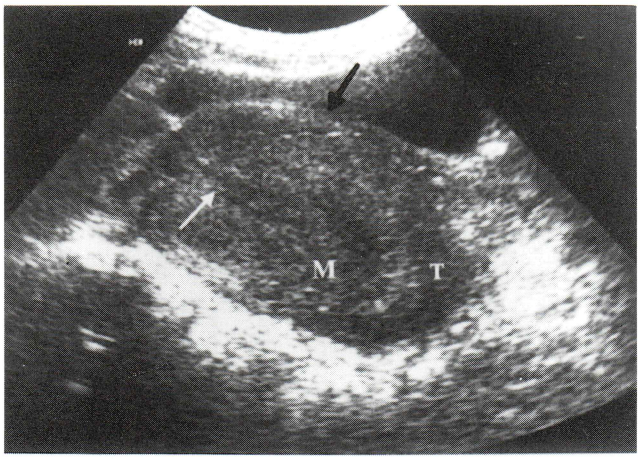


Fig. 1. Enhanced axial scan of the pelvis.

Level just above acetabular roof level. An inverted uterus (U) is demonstrated as a layer of enhancing soft tissue mass located centrally. It is surrounded by hypodense fluid filling the fornix and uterine cavity. The outermost layer represents the dilated endocervix (arrow). A well-defined enhancing soft tissue tumor (arrowhead) is seen posterior to the uterus.



↑ A B →

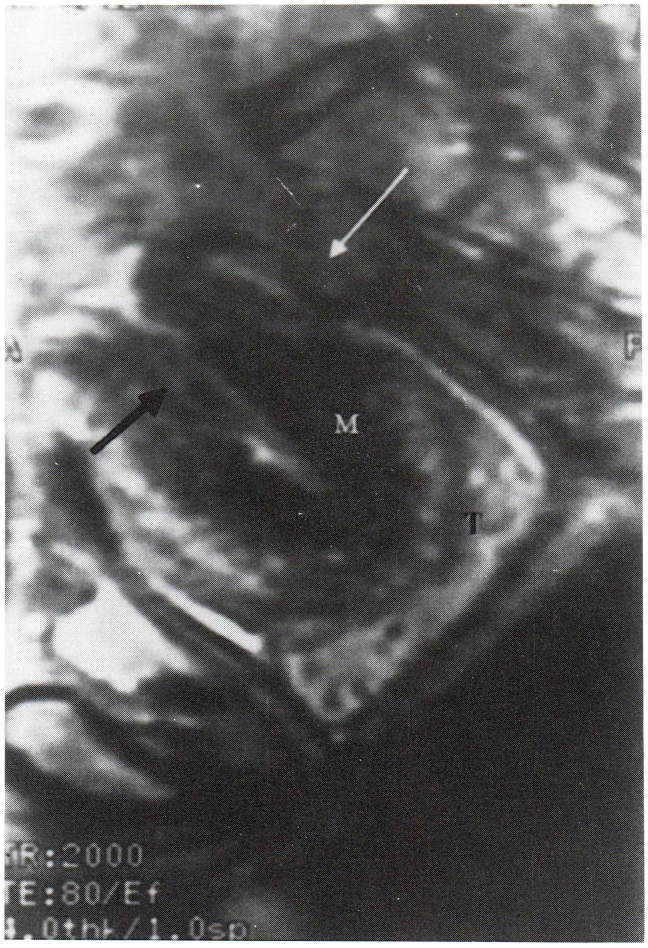


Fig. 2. Parasagittal imaging of the uterine inversion.
A: Parasagittal pelvic ultrasound.
B: MRI T2 weighted.

Both showed an inverted U-shape uterus, which invaginated through the endocervix (black arrow in A and white arrow in B) into vaginal canal. The layer of the uterus from outer to inner, endometrium, myometrium (M), and serosa (arrow), respectively. At the fundus, ill defined tumor mass (T) is seen.

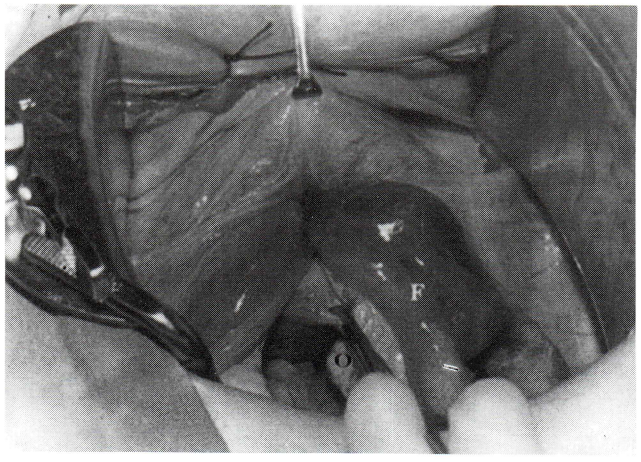


Fig. 3. Completed uterine inversion with round ligaments and some part of the fallopian tubes (F) embeded in the inversion. O:ovary

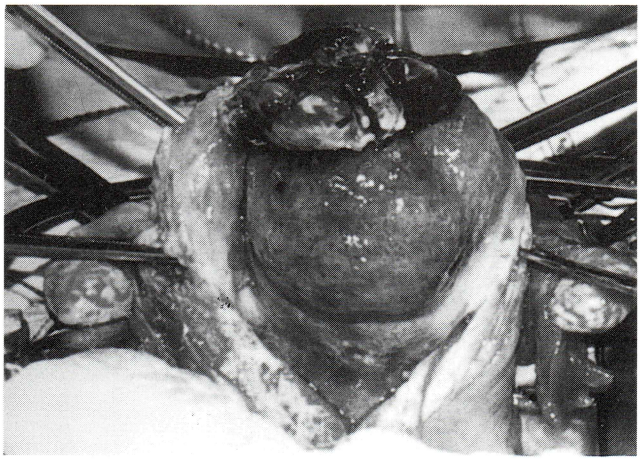


Fig. 4. The inverted uterus with an exophytic mass covered with necrotic tissue after Haultain procedure (performing longitudinal incision along the lower posterior uterine wall)

Discussion

The mechanisms of tumor producing non-puerperal uterine inversion may be from the distended myometrium by the tumor that becomes irritated and develops expulsive contractions, then the cervix is dilated and facilitate expulsion of the tumor and when the tumor passes through the cervical canal, it exerts traction on the weak portion of the myometrium where it is attached, thereby expediting inversion. Additional possible etiologic factors are the weight of the intrauterine mass, manual traction on the tumor, straining, coughing, or sneezing.[2] In our case the tumor was confined to the fundus and motorcycle riding might be the precipitating cause.

Patients with uterine inversion usually have some vaginal discharge or irregular vaginal bleeding, with pelvic discomfort, or pressure in the vagina.[2] It may complicated by infection, hemorrhage, pulmonary embolization or compression of the ureters.[4] On examination, an infected haemorrhagic mass is usually found in the vagina and is likely to be confused with an ulcerated prolapse cervix and even a malignant neoplasm such as cervical cancer[6,7] like our case. Uterine inversion may be misdiagnosed as prolapsed intrauterine tumor and excisional biopsy resulted in severing fundus of the uterus and inadvertent entry into the peritoneal cavity has been reported.[2] Three major points should be kept in mind while performing pelvic examination for this rare condition. First, the cervix may or may not be recognized. Second, the cervical os and endometrium cannot be identified. Third, the uterus is not in its normal position in the pelvis and cupping of the fundus can sometimes be palpated.[2]

Investigations that confirm the uterine inversion are CT-scan,[7] ultrasonography[8] and MRI,[9] which was possibly the best one,[7] as shown in our case.

The diagnosis of this condition should be çkept in mindé and malignancy should be ruled out by performing biopsy. If malignancy is diagnosed, the treatment in this condition is identical to cancer therapy.[10] If the tumor is benign, conservative surgery may be proposed[10] after the complication has been treated.[2]

Four surgical procedures are described to correct the inverted uterus, two via vagina exposure and the other two via abdominal exposure.[2,8,12] The Kustner procedures involves reinversion of the uterus by entering the space of Douglas through the vagina and splitting the posterior wall of the uterus. The uterus can be reinverted into its anatomic position by upward pressure on the fundus. The splitted posterior aspect of the uterus and cervix facilitates this maneuver. The Spinelli operation is similar in the principle, except that the incision into the uterine wall is made anteriorly after the bladder has been retracted upwards. The uterus is then reinverted manually as in the Kustner operation after the incision has been made. Following reposition by either method, the reinverted uterus can be restored by suturing of the line of incision, or vaginal hysterectomy can be accomplished with the uterus in its anatomic position. The Huntington operation consists of making an abdominal incision and grasping the uterus with Allis clamps just below the inversion cup. The cup is pulled up and another pair of Allis clamps grasp the uterus at a lower level; this procedure is repeated until reinversion is completed. The Haultain procedure, also performed by the abdominal route, consists of incision of the constricting cervical ring posteriorly and reinversion of the uterus by traction on the fundus.[2,12] Due to large tumor, our patient underwent TAH & BSO after successful reinversion of the uterus by Haultain procedure. Low grade endometrial stromal sarcoma is a malignant tumor, but local recurrence or distant metastasis may occur many years after initial surgery, especially in case of tumor limited to the uterine corpus like in our case. TAH&BSO is the standard procedure for this condition.[13] However, the patient needs long term follow-up.

In conclusion, non-puerperal uterine inversion is an extremely rare condition, but should be kept in mind. This condition is sometimes related to malignancy and biopsy should be done before offering definite treatment.

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