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An exceptional uterine inversion in a virgo patient affected by submucosal leiomyoma: Case report and review of the literature

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Abstract

Inversion of the uterus is quite a rare obstetric (puerperal) or gynecological (non-puerperal) condition. Non-puerperal type may be benign or malign mass induced or idiopathic. To the best of our knowledge, this is the first case with a large review of literature of uterine inversion in a patient without sexual experience. We report the case of a 52-year-old, unmarried virgo woman, affected by schizophrenia, was referred to our emergency room for evidence of a mass outside of the vaginal introitus, combined with vaginal bleeding and abdominal pain. The patient underwent vaginal myoma resection and abdominal hysterectomy with bilateral adnexectomy. The diagnosis of uterine inversion was confirmed during operation. Diagnosis of uterine inversion is often not easy and imaging studies might be helpful. Despite the rarity, uterine non-puerperal inversion is possible: for this reason, it is necessary to perform appropriate, conservative treatment, especially in women affected by benign myomas and with a desire for pregnancy.

Key words: leiomyoma, non-puerperal uterine inversion, Virgo patient.

Introduction

Uterine inversion is defined as invagination of the fundus by *glove finger* exteriorization up to the vulva. This rare condition was classified by Jones in 1951 into two types: 1) puerperal or obstetric and 2) non-puerperal or gynecologic.[1] Most cases of uterine inversion are puerperal inversions, whereas non-puerperal inversions are rare, and its incidence has not been estimated in the literature.[2] Non-puerperal uterine inversion is an uncommon complication of a benign or malignant uterine mass; it is commonly caused by submucous fundal leiomyomas. In this paper, we report the case of a 52-year-old, unmarried woman diagnosed with a non-puerperal uterine inversion induced by a submucous myoma, managed with vaginal myoma resection and abdominal subtotal hysterectomy.

Case Report

A 52-year-old unmarried woman, Virgo, was referred to our emergency room for suspect uterine inversion. She suffered from schizophrenia. Her surgical history was uneventful. Her legal guardians reported that the patient had been suffering from menometrorrhagia and dysmenorrhea for about 3 years and had already received hysteroscopic and ultrasound diagnosis of a posterior submucous uterine myoma, 4 cm in size, with planned hysteroscopy for a resection. Ovarian tumoral markers tested 2 months previously were negative. Two days before admission in gynecological

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emergency, she had undergone a quadrantectomy for breast Paget disease: during admission there, she complained of a mass protruding from the vaginal introitus, which she tried to push inside manually. After hospital discharge, massive bleeding and abdominal cramping pain occurred and the mass did not diminish. Therefore, she was referred to our hospital. A very large mass, about the size of a fetal-head, protruding from the vagina, with oozing on the surface of the mass, was noted. A uterine inversion with submucous myoma in expulsion was strongly suspected as the uterus could not be identified clearly by transabdominal sonography. Laboratory tests showed moderate anemia (9.6 g/dL). Under endotracheal general anesthesia, the patient was taken up for definitive surgery. A combined abdominovaginal approach was planned (Fig. 1). First, the placement of bilateral ureteral stents was performed. After laparotomic incision according to Pfannenstiel, uterus absence was confirmed, and round ligaments appeared medially stretched. Thus the surgeon first performed a vaginal myomectomy to reduce the volume of the mass protruding from the vagina (Fig. 1a-c). The first attempts to reposition the uterus within the abdominal cavity were unsuccessful: in fact, the Huntington procedure was attempted but failed. Finally, the Haultain procedure was successfully carried out. After that, total hysterectomy and bilateral adnexectomy were performed (Table 1). Following visualization of the ureteral course, colporrhaphy was performed, hemorrhaging was controlled and a pelvic drain was placed. Postoperative evaluation showed well-suspended vaginal wall. Histopathological examination was performed and revealed a leiomyoma with superficial ischemic-hemorrhagic areas, site of hypotrophic endometrium with marked hemorrhagic changes, a polyposis-cystic cervicitis, and scleroatrophic tubes and ovaries.

The postoperative course was smooth, and the patient was discharged and followed up at our outpatients' clinic.

Discussion

We searched electronic medical database in English using keywords as non-puerperal uterine inversion, leiomyoma, uterine surgery. Bibliographies of the relevant articles were reviewed and then cross-searched to identify further relevant studies. Articles that

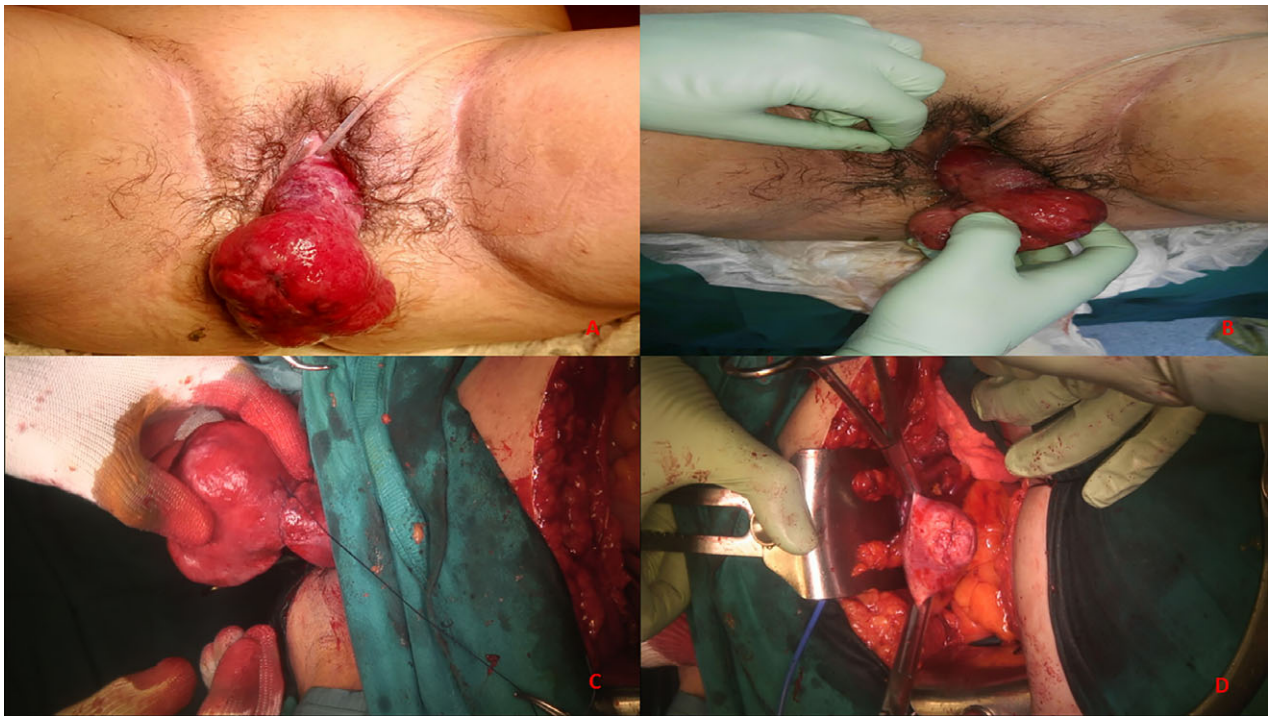

Figure 1 (a-c) The inverted uterus with myoma, seen from vaginal route. (d) The reinverted uterus after myomectomy, seen from abdominal route.

Table 1 Case review of the literature

References	Age	Parity	Previous uterine surgery	Cause of uterine inversion	Mass size	Type of surgery
Our Case	52	V	None	Submucous myoma	4 cm	Combined vaginal/abdominal total hysterectomy and bilateral salphingoopherectomy
Kopal *et al.*[3]	72	MP	None	Two myomas: pedunculated (1) and submucous (2)	13 × 15 cm (1); 5.4 × 5.3 cm (2)	Total abdominal hysterectomy
Hanprasertpong *et al.*[4]	45	NP	None (previous external radiation plus intracavitary radiotherapy for squamous cell carcinoma of the cervix FIGO stage IIa)	Malignant mixed müllerian tumor	10 × 8 × 7.5 cm	Excision of the tumor mass by vaginal route before hysterectomy and bilateral salpingo-oophorectomy by the abdominovaginal approach
Lupovitch *et al.*[5]	26	NP	None	Sarcoma	5 cm	Radical abdominal hysterectomy
Case *et al.*[6]	21	NP	None	Rhabdomyosarcoma	6.5 × 6.5 × 5.7 cm	Total abdominal hysterectomy and bilateral salpingo-oophorectomy
Rosales Aujang *et al.*[7]	46	MP	None	Myoma	NR	Total abdominal hysterectomy
Chen *et al.*[8]	42	V	None	Submucous myoma	15 × 10 × 8 cm	Subtotal hysterectomy
Sharma *et al.*[9]	18	NR	None	Rhabdomyosarcoma	NR	Total abdominal hysterectomy and bilateral salpingo-oophorectomy
Ojwang *et al.*[10]	16	NP	None	Embryonal rhabdomyosarcoma (sarcoma botyroides)	NR	Total abdominal hysterectomy
Pandit [11]	57	NR	None	Sarcoma	NR	Total abdominal hysterectomy and bilateral salphingoopherectomy
Ueda *et al.*[12]	28	NP	D&C	Cervical adenocarcinoma	5 cm	Radical hysterectomy, bilateral salpingo-oophorectomy and pelvic lymphadenectomy
Gomez-Lobo *et al.*[2]	15	NP*	None	Teratoma	2.9 cm	Uterine-sparing surgery
da Silva *et al.*[13]	15	NP	None	Embryonal rhabdomyosarcoma	NR	Total abdominal hysterectomy
De Vries and Perquin[14]	19	NP	None	Submucous myoma	5 cm	Laparoscopy followed by uterine-sparing laparotomy
Fofie and Baffoe[15]	42	MP	None	Myoma	NR	Vaginal hysterectomy
Auber *et al.*[16]	40	MP	Three hysteroscopic resections of submucous leiomyomas	Submucous myoma	7 cm	Combined laparoscopic and vaginal total hysterectomy
Pellissier-Komorek *et al.*[17]	57	MP	None	Submucous myoma	10 cm	Combined laparoscopic and vaginal total hysterectomy
Mehra *et al.*[18]	60	NR	None	Sarcoma	18 × 12 × 6 cm	Total abdominal hysterectomy and bilateral salphingoopherectomy
Shivanagappa *et al.*[19]	28	NP	None	Myoma	12 × 8 cm	Combined vaginal/abdominal total hysterectomy
Umeononihu *et al.*[20]	51	MP	None	Myoma	7.5 × 6.9 × 9 cm	

Table 1 Continued

References	Age	Parity	Previous uterine surgery	Cause of uterine inversion	Mass size	Type of surgery
						Myomectomy followed by subtotal hysterectomy 2 weeks later
Sakıncı *et al.*[21]	39	MP		None	Three submucous myomas	4 × 2 cm (1); 3 × 2 cm (2); 2 × 2 cm (3)
Myomectomy with uterine-sparing laparotomy						
Kouamé *et al.*[22]	28	NR	NR	Submucous myoma	10 × 15 cm	Combined vaginal/abdominal total hysterectomy
Kirbas *et al.*[23]	NR	NR	NR	Submucous myoma	NR	NR

D&C, dilation and curettage; MP, multiparous; NP, nulliparous; NP*, 1 previous spontaneous abortion; NR, not reported; V, Virgo.

provided various diagnostic options and management strategies of non-puerperal uterine inversion were included.

A total of 22 cases of non-puerperal uterine inversion were reviewed (Table 1). Most authors resort to a combined vaginal and abdominal approach to first remove the cause of inversion, and then proceed to the total or subtotal removal of the uterus, but this depends on patient's age and on malignancy of lesion (Table 1). The incidence of uterine inversion, which is one of the extremely rare entities in gynecology, is still not known. There are two types of uterine inversions: puerperal or obstetric and non-puerperal or gynecologic.[1] A uterine inversion is a rare complication of the puerperium, and non-puerperal inversion is an extremely rare occurrence. Gomez-Lobo *et al*.[2] reported 150 cases of non-puerperal uterine inversions documented from 1887 to 2006. Generally, non-puerperal uterine inversion presents after 45 years and is mostly related to benign myomas and seldom associated with malignancies. The cause of uterine inversion is unknown: thin uterine wall, co-existent and rapidly growing tumor, fundal tumor location, tumor with a thin pedicle, size of tumor, dilatation of the cervix by distension of the uterine cavity and a sudden expulsion of the tumor through the vagina have been proposed as etiologic factors.[24,25] Mwinyoglee *et al*.[26] reported that 97.4% of uterine inversions are associated with tumors, out of which 20% were malignant. The most common tumor in the benign group is myoma. On the other hand, the most common tumors in the malignant group are sarcoma, endometrial carcinoma and malignant mixed Mullerian tumor. In idiopathic cases, the condition is usually abrupt, accompanied by pain and shock, while in tumor-produced cases, it is mostly associated with pelvic pain, a mass protruding from the vaginal introitus, urinary disturbance, chronic vaginal discharge, and irregular vaginal bleeding.[1,4] Increase in vaginal discharge, irregular bleeding and pelvic pain are the most common symptoms. Uterine inversion is classified as acute or chronic and subclassified as incomplete or partial (fundus protrudes into the uterine cavity but not through the external cervical os), complete (fundus protrudes through the external cervical os), or complete with inversion of the vagina. Clinical diagnosis of non-puerperal uterine inversion is more difficult, especially when the uterine fundus cannot be palpated. Symptoms may include dysuria and urgency. The openings of Fallopian tubes can be recognized if they have been dragged through the

endometrial surface. If the uterine inversion is complete, a mass protruding from the vulva, with the cervical ring constricting superiorly, may be identifiable, and this makes diagnosis easier. In all other cases, this could be very difficult, especially for partial inversion. In this case, ultrasonography or radiologic exams could be useful. Computed tomography (CT) diagnosis may be difficult, but the absence of the uterus in the pelvis and the visualization of edematous endometrium and myometrium in the middle of the pelvis are suggestive of the condition. Magnetic resonance imaging (MRI) may show the anatomic anomalies more accurately than CT. On MRI scans, a U-shaped uterine cavity and a thickened and inverted uterine fundus are the signs of uterine inversion.[4,27] The role of CT/MRI in these cases is generally limited to chronic cases, as they are helpful in the surgical management as well as in the identification of other organs, such as the ureters or the bladder, which may be affected by the inversion. Treatment of uterine inversion depends on the preoperative diagnosis, reproductive wish of the patient and acute or chronic condition. Surgery is the treatment of choice in the case of non-puerperal inversion. The repositioning of the uterus, respecting the exact anatomy, is vital before proceeding to hysterectomy. It would not be possible to separate and push down the bladder or clamp the uterine vessels for hysterectomy before repositioning the uterus in its proper position. In chronic inversion, surgery is generally necessary, unlike the acute condition, where manual repositioning is possible. Four surgical procedures (two by the vaginal route, Spinelli and Kustner procedure, and two by the abdominal route, Huntington and Haultain procedure) have been used to reinvert the uterus to its correct anatomic position. The Spinelli procedure consists of the anterior approach, with dissection of bladder and uterine anterior wall incision, while Kushner is a posterior approach, with incision on the posterior wall of the uterus. Surgical repositioning of the uterus can also be done through the abdominal route, such as the Huntington approach, which consists of locating the uterus cup, dilating the cervical ring manually and gentle upward traction of round ligaments. The Haultain procedure employs a vertical incision on the posterior portion of the cervical ring and gentle traction on the round ligaments.[24,25,27] A review of the relevant literature suggests that most authors resort to a combined vaginal and abdominal approach to first remove the cause of inversion (in more than 80% of cases a submucosal myoma),[4] and then proceed to the total or subtotal removal of the uterus, associated or less at bilateral salphingoopherectomy, generally depending on patient's age and on malignancy of lesion, as reported in Table 1. We found that four cases of sarcoma induced uterine inversion, and Case[6], Pandit[11] and Mehra et al.[28] opted for total abdominal hysterectomy and bilateral salphingoopherectomy.[4] Hanprasertpong *et al*.[4] performed the excision of the tumor mass by vaginal route before hysterectomy and bilateral salpingo-oophorectomy by the abdominovaginal approach because of malignant mixed müllerian tumor or carcinosarcoma (Table 1). Fofie and Baffoe[15] performed a vaginal hysterectomy in multiparous 42-year-old women affected by a submucosal myoma. Chen *et al*.[8] described, for women with the same age and benign pathology, a subtotal hysterectomy, after vaginal removal of mass. This case is similar to ours due to the fact that the patient was without sexual experience. However, our case is differentiated by the fact that the patient was affected by schizophrenia, and this may have led to a delay in the diagnosis, as, during the days before hospitalization, the woman tried to manually reduce a mass protruding from the vagina, without requiring medical evaluation, despite being admitted to another hospital for treatment of breast Paget. This may have transformed a partial chronic inversion into an acute inversion, requiring a more destructive treatment.

In literature, the youngest patient affected by uterine inversion was a 19-year-old nulliparous woman with abnormal vaginal bleeding, dysmenorrhea, and a large mass protruding from the cervix.[14] Only in one case, non-puerperal uterine inversion was associated with an immature teratoma of the uterus in a 15-year-old patient with a history of a previous spontaneous abortion 6 months before her hospitalization.[2] Three teenagers, aged 15, 16 and 18 years, were diagnosed with rhabdomyosarcoma,[9,10,13] and Ueda *et al*.[12] reported a uterine inversion in a 28-year-old diagnosed with endometrial cancer (Table 1).

Therefore, for women with benign disease abdominal or vaginal hysterectomy is recommended, especially for women who do not desire any (further) pregnancy. A more conservative treatment (subtotal hysterectomy or uterine-sparing approach) could be performed for benign disease in the other cases of clinically stable women. When a uterine malignancy is associated with uterine inversion, abdominal hysterectomy with appropriate staging surgery is usually indicated. From the available literature, a vaginal-

laparoscopic combined approach for hysterectomy is now emerging.[3,5–7,16,17,19–23]

A case of non-puerperal uterine inversion due to benign submucous myoma has been reported. This finding seems to be very rare in the literature considering that the woman had already noticed the presence of the mass protruding from the vagina, but she had not asked for help, causing a chronic partial inversion to become acute. Based on the literature data, it seems that mass size is not a relevant risk factor for the uterine inversion, considering that it could be caused both by a small lesion of 2 cm and one of 18 cm. Despite the rarity, uterine non-puerperal inversion should be considered possible and avoided in all those predisposing conditions, such as submucous myomas, especially in women with a desire for pregnancy, promptly resolving the pathology in progress. Uterine-sparing surgery should be attempted in young women, until the final pathology of the disorder is known. In chronic conditions, ultrasonographic or radiographic markers of partial inversion should be identified, so as to stop the inversion process in time and allow a correct differential diagnosis. In these cases, histopathologic evaluation is necessary to exclude malignant causes. Also, examination under anesthesia could be useful in all those cases with diagnostic doubts.

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