

CASE REPORT

Gynecology

Nonpuerperal uterine inversion with a submucosal leiomyoma: A case report

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Abstract

Background: Nonpuerperal uterine inversion is a rare gynecological condition that is not preceded by pregnancy. It may be induced by uterine tumors or may be idiopathic.

Case presentation: A 28-year-old presented with a vaginal mass associated with urine retention, vaginal bleeding, and an off-white discharge. Pelvic sonography performed earlier revealed uterine procidentia features. On examination under anesthesia, the vaginal mass turned out to be an inverted uterus with a submucosal fibroid. The fibroid was avulsed. Bladder dissection and hysterotomy were performed, and the uterus was reduced into the

abdominal cavity. She recovered well postoperatively with complete symptom resolution.

Conclusion: Nonpuerperal uterine inversion is a rare and potentially life-threatening condition, which presents with nonspecific symptoms and presents diagnostic and management challenges. Management is usually surgical as conservative measures have been shown to be less effective. Timely diagnosis and management is key in reducing associated morbidity and mortality.

Keywords: leiomyoma, nonpuerperal, uterine inversion, vaginal reduction

Introduction

Nonpuerperal uterine inversion is a rare gynecological condition that is not preceded by pregnancy. Its incidence remains unclear with about 170 cases reported in literature (1). It is often preceded by uterine tumors which trigger myometrial distension and irritability resulting in expulsive contractions and cervical dilatation (2). The commonly associated symptoms are a protruding vaginal mass, pelvic pain, irregular vaginal bleeding, and vaginal discomfort. Symptoms may be acute or chronic with an insidious inception (1). The diagnosis is often difficult and imaging modalities, such as pelvic sonography and magnetic resonance imaging (MRI) are helpful (2). Examination under

anesthesia (EUA) and laparoscopy are key to confirm the diagnosis. There is no clearly established protocol for management and surgical reduction and reinstitution are the typical objectives. This is influenced by the underlying pathology and fertility desires of the patient (3). This is a case of nonpuerperal uterine inversion associated with a submucosal leiomyoma.

Case presentation

A 28-year-old para 1 gravida 1 whose last delivery was 9 years prior presented to the urogynecology clinic at Kenyatta National Hospital (KNH). She had experienced sudden profuse vaginal bleeding that was preceded by an off-white discharge associated with backache and suprapubic pain for

more than a month. She also felt a protruding vaginal mass with difficulty in voiding. A diagnosis of prolapsed uterine fibroid was made at a nearby health facility after a biopsy was taken. She was managed conservatively with antibiotics and analgesics for two weeks and discharged with a urethral catheter pending reduction. Subsequently, a pelvic ultrasound scan done was suggestive of uterine procidentia. Manual reduction was attempted unsuccessfully and a referral to KNH was made.

At KNH, an inverted uterus with a submucosal fibroid was noted during EUA (Figure 1). Tew's technique was used to achieve reduction and restitution of the uterus. This procedure involved avulsion of the submucosal fibroid and an anterior hysterotomy from the fundus to the level of the cervix exposing the fallopian tubes and round ligaments (Figure 2). The bladder was dissected off the cervix and the hysterotomy incision was partially sutured after rotating the uterus inside out (Figure 3). The uterus was then reduced to its anatomical position and the rest of the hysterotomy incision was sutured (Figure 4). A drain was placed in the vesicouterine space to prevent hematoma formation and a vaginal pack was left for 24 hours. The drain was removed after 48 hours, and the patient was discharged on the third postoperative day. The patient's menstrual bleeding resumed two weeks postoperatively and she was complication-free three months following surgery. She was counselled on the need for follow up in the high-risk antenatal clinic in subsequent pregnancies and the recommendation to have elective caesarian section at her next birth given the extensive uterine upper segment surgery that was performed.

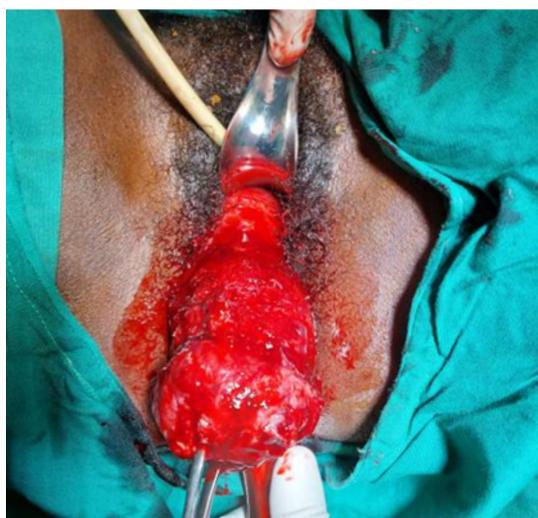


Figure 1: Inverted uterus with fundal submucosal fibroid.

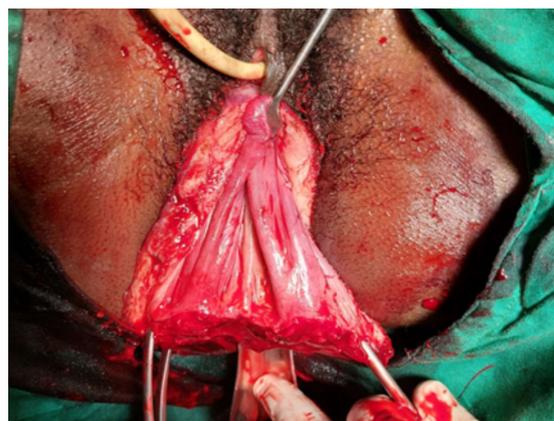


Figure 2: Anterior uterine hysterotomy exposing the round ligaments and fallopian tubes.



Figure 3: Partially sutured uterine incision prior to reversion of uterus.

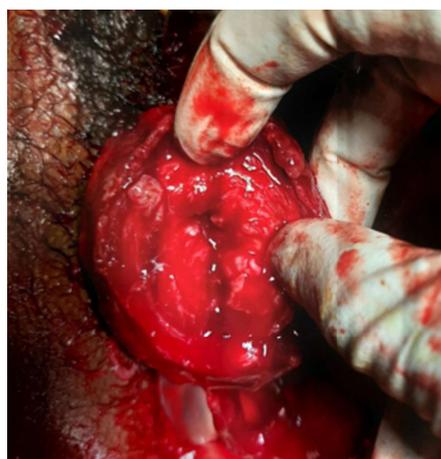


Figure 4: Visualization of the cervix after uterine reversion.

Discussion

Uterine inversion is the turning inside out with subsequent prolapse of the uterine fundus into the uterine cavity or the vaginal canal. It can be classified as puerperal occurring after delivery or

nonpuerperal occurring remotely from pregnancy (4). Puerperal uterine inversion is fairly common occurring mostly due to inappropriate obstetric practices (5). Nonpuerperal uterine inversion is a much rarer entity that presents diagnostic and management challenges. It is often preceded by uterine tumors which trigger myometrial distension and irritability resulting in expulsive contractions and cervical dilatation (2). These tumors may include submucosal uterine fibroids, endometrial polyps, uterine sarcomas, and endometrial carcinomas. About 60% of nonpuerperal uterine inversion cases have been shown to be associated with uterine leiomyoma, which are common in African women as in this case (1). Other factors predisposing to nonpuerperal uterine inversion include fundal location of the tumor, large tumor size, large tumor pedicle, and thinness of the uterine wall (7). Acute cases of uterine inversion may present with excruciating pain and hemorrhage while chronic cases may present with pelvic discomfort, irregular vaginal bleeding, anemia, and vaginal discharge (5). Mortality due to uterine inversion is approximately 15% (6). Here, the patient presented with sudden profuse vaginal bleeding, vaginal discharge, and a protruding vaginal mass associated with urine retention.

Diagnosis of the nonpuerperal uterine inversion can be challenging given the nonspecific symptomatology (3). Diagnosis is usually easier for the higher grades of inversion extending beyond the introitus where a bluish-red mass may be identified on examination. In challenging cases, imaging studies like pelvic sonography and MRI may be used (2). Here, the diagnosis was missed at lower-level healthcare facilities and was only made intraoperatively at the national hospital. The choice of surgical procedure between hysterectomy and uterine reduction for inversion depends on the patient's fertility desires and the surgeon's experience (3). Manual reduction and hydrostatic reduction usually used in puerperal uterine inversion may not be successful in the nonpuerperal inversion due to the gradual contraction of the cervix and lower uterine segment to create a constriction ring (3,4). Several surgical approaches to uterine reduction have been described including vaginal, abdominal, laparoscopic, and robotic.

Two abdominal approaches, Haultain and Huntington have been described in literature. In the Haultain procedure, a longitudinal incision is made in the posterior uterine wall extending into

the constriction ring and traction is applied on the round ligaments to achieve the reduction (8). The Huntington procedure can also be used in which case the cup of the uterus caused by the inversion is identified followed by digital dilatation of the cervical ring. The round ligaments are then located and gentle traction is applied onto them to complete the reduction (9). Transvaginal reduction has been elaborately described using Tew's and Kustner's techniques. In Tew's technique, the bladder is dissected off the cervix and lower uterine segment. A longitudinal incision is then made on the anterior uterine wall to release the constriction ring. The uterus is reverted and the hysterotomy incision is repaired in layers (3). This uterine conserving technique was used in this patient's case since she had fertility desires. In Kustner's technique, an incision is made in the posterior cul-de-sac, cervix, and posterior uterine wall. The uterine fundus is then reduced through the incision prior to repairing the hysterotomy incision and the cervix. Finally, the colpotomy incision is repaired (10). Due to rarity of the condition, successful outcome is highly dependent on surgical proficiency in gynecological procedures. In this case, a urogynecologist was involved.

Conclusion

Nonpuerperal uterine inversion is a rare and potentially life-threatening condition, which presents with nonspecific symptoms and presents diagnostic and management challenges. Management is usually surgical as conservative measures have been shown to be less effective. Timely diagnosis and management is key in reducing associated morbidity and mortality.

Consent for publication

Informed consent for publication was obtained from the patient.

Acknowledgment

Not applicable.

Declarations

Conflict of interests

The authors declare no conflicts of interest.

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References

1. Rosa Silva B, de Oliveira Meller F, Uggioni ML, et al. Non-puerperal uterine inversion: a systematic review. *Gynecol Obstet Investig.* 2018;83:428-436
2. Herath RP, Patabendige M, Rashid M, Wijesinghe PS. Nonpuerperal Uterine Inversion: What the Gynaecologists Need to Know?. *Obstet Gynecol Int.* 2020;2020:8625186. Published 2020 Jun 1. doi:10.1155/2020/8625186
3. Pinder LF, Ouma KO, Nelson BD. Non-Puerperal Uterine Inversion in a Young Woman: A Case Report, Brief Surgical Review, and Clinical Insights. *Clin Med Rev Case Rep.* 2016;3(8):122
4. Ward HR. O'Sullivan's hydrostatic reduction of an inverted uterus: sonar sequence recorded. *Ultrasound Obstet Gynecol.* 1998;12(4):283-286. doi:10.1046/j.1469-0705.1998.12040283.x
5. Ymele FF, Nana P, Fouedjio JH, Bechem R E, Mbu R. Non-puerperal uterine inversion following a prolapsed leiomyoma in a Cameroonian woman. *Clinics in Mother and Child Health.* 2011;8:1-3
6. Minakshi S, Shivani A, Arshad A. Neglected puerperal inversion of the uterus: ignorance makes acute a chronic form. *Pan Afr Med J.* 2012;12:89
7. Fofie CO, Baffoe P. Non-puerperal uterine inversion: a case report. *Ghana Med J.* 2010;44(2):79-81. doi:10.4314/gmj.v44i2.68889
8. Haultain F. W. N. The treatment of chronic uterine inversion by abdominal hysterotomy, with a successful case. *Br Med J.* 1901;2(2127):974-976
9. Huntington JL, Irving FC, Kellogg FS. Abdominal reposition in acute inversion of the puerperal uterus. *Am J Obstet Gynecol* 1928; 15:34
10. Birge O, Tekin B, Merdin A, Coban O, Arslan D. Chronic Total Uterine Inversion in a Young Adult Patient. *Am J Case Rep.* 2015;16:756-759. Published 2015 Oct 23. doi:10.12659/ajcr.894264