



SUBMUCOUSAL FUNDAL FIBROID CAUSING INVERSION OF UTERUS: A CASE REPORT AND LITERATURE REVIEW

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ABSTRACT

Inversion of the uterus is an uncommon complication of the puerperium and it is an even rarer complication of the non-puerperal period. A submucous myoma is mostly the cause of the non-puerperal inversion.

As nonpuerperal uterine inversion is rarely encountered by the gynecologist and, the fact that many gynaecologists might not see any in their entire practice gives a clue as to its rare occurrence, thus diagnosis and management remains challenging. This requires high index of clinical suspicion for its proper diagnosis. The case presented here was a 42 years old para four lady with complaints of severe back ache and pain abdomen since 4 days, some discomfort in vagina and profuse bleeding per vagina since 2 days. She was referred from local hospital as a case of incomplete abortion. Pelvic ultrasound suggested the diagnosis of uterine inversion due to fundal submucosal fibroid. She was managed by Huntington procedure to reposition the uterus followed by total abdominal hysterectomy with bilateral salphingoophorectomy. This article is expected to increase awareness of the confusing clinical presentations of this “gynaecological near miss” entity.

Key Words: Non-puerperal; chronic; uterine inversion; fundal submucosal fibroid

INTRODUCTION

Uterine inversion refers to a descent of the uterine fundus to or through the cervix, so that the uterus is turned inside out. [1,2] Uterine inversion occurs when the uterine fundus prolapses into the endometrial cavity, with resultant partial or complete inversion exposing endometrial surface. [1,3,4] The openings of the fallopian tubes may be identifiable if it had been dragged through the endometrial surface. [1] It is categorized as puerperal uterine inversion when it occurs in the postpartum period and non-puerperal uterine inversion when it occurs secondary to benign or malignant uterine masses in non-pregnant women. [1,3,5]

Non-puerperal uterine inversion is extremely rare with no specific reported incidence, though it forms one-sixth of all inversions. [2,3,5] Although non-puerperal inversion is often chronic, it has been reported that 8.6% of non-puerperal inversions may have a sudden onset [3,4,6]. It is often associated with the presence of a polypoid uterine tumor. Mwinyoglee et al. reported that 97.4% of uterine inversions were associated with tumors, of which 20% were malignant, while Takano et al. reported that 71.6% of cases of uterine inversion were associated with leiomyoma's. [3,6,7].

Most non-puerperal inversions are caused by benign submucous myomas, while other causes are leiomyosarcoma, rhabdomyosarcoma, malignant mixed müllerian tumour and endometrial polyp. Women of reproductive age who present with the rare finding of non-puerperal uterine inversion are likely to have a malignancy. [5,7,8]

The aetiology of uterine inversion is not clearly defined. Possible explanations could be a thin uterine wall, rapid growth of the tumour, tumour size, fundic localisation of the tumour, tumour attachment to the uterine wall with a thin pedicle, dilatation of the cervix by distension of the uterine cavity, and sudden expulsion of the tumour [5] Non Puerperal inversion of uterus can be caused by a pedunculated tumor attached at the fundus or by leiomyoma of uterus. Sessile fundal fibroid causing uterine inversion has either not been individually cited or no particular reference has been made. The fundal fibroid causes thinning and weakness of uterine wall due to pressure atrophy. Contraction of uterine musculature excited by the prolapse of the tumor into the cavity causes inversion of uterus. A fundal fibroid can cause uterine inversion only if it is sufficiently large and heavy. This process takes years making uterine inversion due to myoma a rare possibility simply because submucosal myomas produce a variety of symptoms such as menorrhagia or vaginal discharge before the occurrence of inversion and women usually seek help for various reasons, quite early in the course [6].

Once the tumour descends into the uterine cavity, uterus tend to expel the submucous myoma with fundal attachment which ultimately leads to inversion. This is most likely responsible for the symptom like lower abdominal pain and backache. [5]

Prolapsed fibroids tend to be the most common inciting factor with occasional reports of inversion associated with uterine neoplasm and endometrial polyps.

Three striking features proposed for uterine inversion are: 1) sudden emptying of the uterus which was previously distended by a tumor 2) thinning of the uterine walls due to an intrauterine tumor, and 3) dilatation of the cervix. [9]

Uterine inversion can be classified into four stages as Stage 1: The inverted uterus remains in the uterine cavity, Stage 2: Complete inversion of the fundus through the cervix, Stage 3: The inverted fundus protrudes through vulva, and Stage 4: Inversion of the uterus and vaginal wall through the vulva. Inversion can also be classified as acute and chronic. With acute inversion, the patient may have severe pain in lower abdomen or excessive bleeding whereas chronic inversion may be insidious or patient may have lower abdominal discomfort, vaginal discharge, irregular vaginal bleeding, or anemia. In chronic inversion with sloughing of endometrium diagnosis, it is not very easy so detailed abdominal, and vaginal USG or Doppler may be required to confirm the diagnosis. [2,6]

Acute uterine inversion is an un-common complication of parturition which often occurs in the immediate postpartum period, it is attributed to mismanagement of third stage of labour and is an acute obstetric emergency. [1,9] --ab] Puerperal inversion occurs in 1: 3500 deliveries. [6] It causes severe pain and haemorrhage often leading to shock. [1]

Chronic uterine inversion is defined as acute inversion which has been present for at least 4-weeks after delivery and which has resisted the standard methods for reposition. It may follow puerperal or obstetric cases which have been overlooked, or those associated with gynaecologic cause, like the extrusion of tumours of the uterine fundus. It is a very rare condition and an incidence of 1 in 8500 has been reported in resource-poor countries. [7]

Uterine inversions is a rare condition and most of it are acute and puerperal (85.8%). Non-puerperal inversions are described in very few patients and are also said to be chronic or gynecological uterine inversion. They represent about one sixth of all inversion cases (16.35%). Non-puerperal inversions occurring acutely is rare and so far there was report in which 8.6% of the non-puerperal uterine inversion occurred suddenly. [4] Non-puerperal uterine inversion has been described as a rare occurrence by several authors but how really rare is it has not been defined. However, the fact that many gynaecologists might not see any in their entire practice gives a clue as to its rare occurrence. [7]

Incomplete non-puerperal uterine inversion is a rare complication that can arise secondary to the presence of submucous fibroid. In the majority of cases reported in the literature, the definitive diagnosis is usually made at the time of hysterectomy [10]

Repositioning of the uterus can be done manually in acute cases but in chronic nonpuerperal case, manual reposition is not possible, especially in those cases associated with tumors. In chronic nonpuerperal cases, surgery is imperative. Considering patient's age, reproductive desire and associated conditions, surgical repositioning, or hysterectomy can be done. [1,2]

Surgical procedures described in the literature use different techniques to first reposition the uterus followed by hysterectomy. However, repositioning the uterus is not always successful. Surgery for inverted

uterus is technically difficult due to close proximity of the ureters to the ovarian and uterine vessels due to traction on the vascular pedicles, difficulty in repositioning the uterus and constraints of mobilizing the bladder down due to the inverted uterus. [7]

CASE REPORT

A 42 years woman presented to casualty (of Paropakar Maternity and Women's hospital, Kathmandu, Nepal) with complaints of severe back ache and pain abdomen since 4 days, some discomfort in vagina and profuse bleeding per vagina since 2 days. She was referred from local hospital as a case of incomplete abortion; though urine pregnancy test (UPT) was reported to be negative. On the ground that she was sexually active and was having mild irregular minimal bleeding per vagina with history of passage of clots and fleshy mass occasionally for more than one month. She was married for last 24 years. She had four normal vaginal delivery at home in presence of traditional birth attendants, the last one being 12 years back. There was no prior history of abortion and all the four children were fine. She had been using intrauterine contraceptive device (IUCD) for family planning which was removed 6 months back. To her perception IUCD was causing back ache and increased menstrual flow for the past 1 year. She attended menarchy at the age of 13 years. Previously she was having normal menstruation with regular cycle of 32- 34 days. Her last menstrual period was 6 weeks back since then irregular minimal bleeding per vagina. Her past medical, surgical, and family histories were not of clinical relevance. On examination - she was conscious and irritable, apparently pale, with a pulse rate of 100 beats per minute, temperature 98.8 F, and BP 90 / 60 mmHg. On per abdomen examination - abdomen was soft, mild tenderness in lower abdomen. On other systemic examination -no obvious abnormality was detected.

Per speculum examination showed dilated cervical os with bleeding and beefy red clot like mass in the cervical canal, it was somewhat resembling product of conception on inspection. The patient was screaming on per speculum examination so the bimanual examination could not be done. Blood was sent for complete blood count and urine for routine examination and UPT. She was admitted with provisional diagnosis of incomplete abortion. She was given intravenous fluids for resuscitation, antibiotics and analgesic. Next day morning pelvic examination revealed firm, globular, intracervical mass with distorsion of uterine cavity. Abdominal and pelvic ultrasound was done which suggested the diagnosis of uterine inversion due to fundal submucosal fibroid.

On laboratory investigation important findings were : Blood group was "O" Rhesus -Positive, Hb was 6.4 gm/dL (Normal range: 12-16 g/dL), Hematocrit was 22% (Normal range: 36%- 47%), RBC was 2.3 million/mm³ (Normal range: 4.5-5.5 million/ mm³), WBC was 8000 / μ L (Normal range: 4000-10,000/ μ L), Platelet count was 230,000/ μ L (Normal range: 150,000-350,000/ μ L), Prothrombin time was 10.50s (Normal range: 10-13 s), INR was 1s (Normal range: 0.80-1.20s), serum creatinine was 0.8 (Normal range: 0.7-1.3 mg/dL), Random plasma glucose was 89 mg/dl (Normal range: 70-140 mg/dL). UPT-Negative, urine routine examination showed plenty RBC, few WBC, no bacteria.

After proper resuscitation and transfusion of four units of whole blood, she was prepared for surgery. Intra-operative finding was consistent with partial inversion of uterus with dimpling at center of the upper part of the uterus. Huntington procedure to reposition the uterus followed by total abdominal hysterectomy with bilateral salphingoophorectomy was done. She had smooth recovery and was discharged on sixth post-operative day.

DISCUSSION

Non-puerperal uterine inversion is a rare condition that can pose diagnostic and management dilemmas. [3]

The diagnosis is based on high index of suspicion. [1] Reports have included women of different age groups; the youngest patient was 19 years old with a benign submucous fibroid, and the oldest was 79-year-old with a mixed mullerian tumor. While non-puerperal uterine inversion can occur in both nulliparous and multiparous women, there have been only few reported case in a virgin. [3] The clinical features may include: lower abdominal pain or tenderness, vaginal bleeding, urinary frequency, dysuria and urgency. [1,5] Finding a mass coming through the cervix without definite margins of a cervix, absence of the uterine fundus or fundal dimpling during bimanual or rectal examinations are strongly suggestive of the diagnosis. The openings of the fallopian tubes may be identifiable if it had been dragged through the endometrial surface. [1]

The diagnosis of inversion may be difficult to make during examination. The two findings, namely: (1) the uterine corpus is not palpated on bimanual examination, and (2) the cervix not visualised due to the vaginal mass can be supportive findings. [4,5] The diagnosis is easier with complete inversion when a bluish-red mass is identified from the vulva with a constricting ring of the cervix superiorly. In other cases, the diagnosis can be very difficult. [1,4] Imaging procedures such as ultrasound and magnetic resonance imaging will assist the diagnosis. [1,5] Unfortunately, because of the rare nature of the disorder, uterine inversion frequently goes undetected until surgery unless a high index of suspicion is maintained. [1]

In the majority of cases reported in the literature the definitive diagnosis of uterine inversion was confirmed intraoperatively at the time of hysterectomy [3,5,10] A high index of suspicion with help from radiological tools is required to make the diagnosis and plan management accordingly. [3] Ultrasound examination is the first line imaging investigation. The suggestive features include: indentation of the fundal area and depressed longitudinal groove extending from the uterus to the centre of the inverted uterus. When available, Magnetic Resonance Imaging (MRI) and computerized tomography (CT) scan, are useful diagnostic tools. [1,6] Lewin et al. recommend the use of T2-weighted MRI scans to detect a U-shaped uterine cavity, thickened and inverted uterine fundus on a sagittal image and a 'bulls-eye' configuration on an axial image as two indicative signs of uterine inversion. This however, is not readily available in most hospital settings in developing countries. [3,6,7] Frozen section of the vaginal mass has been used by some clinicians for the diagnosis. Demonstration of the endometrium on the surface of the mass will confirm the diagnosis. Biopsy of the mass has definite place if an associated malignancy is suspected. [1]

Reposition procedures like that of Johnson are more likely to be successful in acute inversion but in chronic cases, surgery is imperative. Depending on the patient's reproductive desire and associated conditions, surgical reposition or hysterectomy could be considered. [7]

Doing a surgery for inverted uterus is challenging, due to the distortion of normal anatomy. It may be feasible to safely accomplish abdominal hysterectomy for uterine inversion without attempting to reposition the uterus. [2,3]

The principle of treatment is to restore the fundus to its correct position by section of constricting ring of the cervix, and then transected cervix to be repaired by suture followed by hysterectomy by vaginal or abdominal approach in multiparous women. It is not desired to conserve uterus in multiparous women. [9] Uterine-sparing surgery should be attempted in young women until the final pathology of the disorder is known. [3,5,8]

Vaginal surgical approaches are rarely performed. Transvaginally, the cervical constriction ring is incised posteriorly (Cascarides procedure) or anteriorly (Spinelli procedure), which should allow the uterus to be repositioned manually. The incision is then repaired. Incidental cystotomy is a hazard of the anterior approach. [9]

Most of the surgical methods described involve reinverting the uterus before both repairing the incisions made and proceeding to hysterectomy or outright hysterectomy. Vaginal hysterectomy without reinverting the uterus has been reported. Repositioning of the uterus is usually done after the tumour has been removed and malignancy must be excluded. Surgery is imperative in chronic inversion unlike in acute inversion where manual repositioning is possible. [1,2]

Spinelli and Kustner are similar trans-vaginal surgical reposition techniques with the basic differences being that Spinelli's approach is anterior and requires dissection of the bladder and an anterior uterine wall incision, while Kustner's is a posterior approach with incision on the posterior uterine wall, which makes it a bit easier and safer. [1]

The Spinelli's operation for chronic inversion of the uterus: The technique involves dissection of the bladder from the inverted uterus. A midline split is made in the cervix and it is carefully separated from the bladder. The anterior wall of the everted uterus is split. By pressure with the operator's index fingers and thumbs the uterus is turned outside in. The myometrium is reapproximated by two layers of running suture, and the serosal surface by a single layer. The vaginal skin is reapproximated with interrupted sutures, as is the full thickness of the cervix. [2,7,8]

The Kustner's operation for chronic inversion of the uterus: After opening the posterior cul-de-sac the cervix and posterior wall of the uterus are incised. On completion of this step, thumb pressure along the sides of the uterus produce reversion, Interrupted sutures are used to close the incisions and the uterus replaced in the pelvic cavity. This is followed by closure of the colpotomy. [1,2,8]

However, surgical repositioning can also be done through a laparotomy using the Huntington procedure, which consists in locating the cup of uterus formed by the inversion, dilating the cervical ring

digitally, and gentle upward traction of the round ligaments of the uterus. [1] Huntington's Approach: In this method pull is applied on round ligaments after laparotomy. Allis forceps is placed on round ligament about 2 cms below the insertion on both sides. Gentle traction is exerted clamps are advanced 2 cms below the previous clamps and the process is repeated till reduction is complete. [2,8]

The Haultain procedure uses a vertical incision in the post portion of the ring and gentle traction on the round ligaments. [1] Haultain's operation Principle: After opening the abdomen the constriction ring is divided posteriorly and the inversion is corrected and incision is closed in two layers. [1,2,8]

We used the Huntington procedure for our patient since it was incomplete inversion and was technically easier for us followed by total abdominal hysterectomy with bilateral salphingo-oophorectomy.

Nevertheless, in places where facility and expertise exist, this repair could be done laparoscopically [1] Laparoscopy reveals the characteristic flower vase appearance of ovaries with no uterus inside the pelvis [8]

Auber et al., described a case of non-puerperal uterine inversion using combined laparoscopic and vaginal approach. This method of approach is now emerging from available literature. [1,8].

Vaginal restoration and removal is difficult. Abdominal hysterectomy with due care to locate the distal ureters along with intraoperative cystoscopy or bilateral ureteric stenting with DJ stent under fluoroscopy guidance to ensure bladder and ureteral integrity is necessary. [3,8]

CONCLUSION

Chronic uterine inversion is a rare condition that is difficult to diagnose even for the experienced gynecologists. On encountering a large prolapsed fibroid one should suspect the presence of chronic nonpuerperal uterine inversion. It is advisable to perform biopsy of the mass in view of its association with uterine malignancy. Uterine inversion has a good prognosis when managed timely in correct manner. The treatment for chronic uterine inversion is surgical that includes both abdominal and vaginal approaches.

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