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## Case Report

# A rare case of acute non puerperal complete uterine inversion in nulliparous virgin female

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## ABSTRACT

Non-puerperal uterine inversion (NPUI) is extremely rare and accounts for 17% of all uterine inversion cases. A few more than 150 cases have been reported amongst which 10-15 have been reported in nulliparous females and 3-4 have been reported in virgin females from 1914 till date. A 40 years unmarried nulliparous female denying sexual activity presented to our hospital with acute non puerperal complete uterine inversion with something coming out per vaginum of sudden onset while straining at defecation, lower abdominal pain and minimal bleeding per vaginum with history of uterine fibroid and chronic constipation. Diagnosis was confirmed with ultrasonography and examination under anaesthesia after written informed valid consent. After detailed counselling and obtaining written informed valid consent, the patient underwent exploration with a combined abdominal-vaginal approach. Following vaginal myomectomy, the uterus was repositioned using Haultain procedure after a failed attempt of Huntington procedure. Total abdominal hysterectomy with bilateral salpingectomy with vaginal vault suspension was done sparing bilateral ovaries. Diagnosis requires a high index of suspicion and their management is a challenge to gynaecologists due to its rare occurrence, distorted pelvic anatomy and associated pelvic organ injuries during surgery. Good anatomical and clinical knowledge along with surgical skills is of utmost importance for successful outcomes.

**Keywords:** Non puerperal uterine inversion, Nulliparous, Virgin, Acute complete uterine inversion

## INTRODUCTION

Uterine inversion refers to descent of the uterine fundus to or through the cervix, so that the uterus is turned inside out.<sup>1</sup> It is an unusual entity and can be classified as puerperal or obstetric and non-puerperal or gynaecological uterine inversion.<sup>2</sup> Sometimes other reproductive structures such as the fallopian tubes and ovaries may also be displaced from the pelvis and confined within the inverted uterus.<sup>3,4</sup> NPUI is extremely rare and accounts for 17% of all uterine inversion cases. A few more than 150 cases have been reported amongst which 10-15 have been reported in nulliparous females and 3-4 have been reported in virgin females from 1914 till date (Table 1).<sup>3,5</sup>

Prolapsed fibroids especially fundal fibroid polyp tends to be the most common aetiology with occasional reports of inversion being associated with uterine neoplasms like

leiomyosarcoma, rhabdomyosarcoma, malignant mixed Mullerian tumour and endometrial polyp.<sup>1,4</sup> They usually present with menorrhagia, mass per vagina and abdominal pain.

**Table 1: Incidence of uterine inversion.**

| Incidence of puerperal uterine inversion | 1 in 20000   |
|--|--|
| Overall incidence of NPUI                | 153 (from 1914 to till date) Herath et al <sup>7</sup>   |
| NPUI in nulliparous women                | 10-15 (from 1914 to till date) Herath et al <sup>7</sup> |
| NPUI in virgin women                     | 3 (from 1914 to till date) Alsahabi et al <sup>8</sup>   |

Diagnosis is based on clinical examination, but confirmation can be done through radiologic

investigations (sonography, MRI, CT) and histopathology.<sup>2,4</sup> In many cases, the diagnosis however is often confirmed during examination under anaesthesia, laparotomy, or laparoscopy.<sup>4</sup> Management and treatment largely depends on the histologic results.<sup>4</sup> Most cases are managed by abdominal or vaginal hysterectomy after repositioning the uterus.<sup>6</sup> Although conservative surgery is preferred among women who wish to preserve fertility, radical hysterectomy is recommended in malignant cases.<sup>4</sup> We report a rare case of non-puerperal complete uterine inversion in nulliparous virgin female and its management.

### CASE REPORT

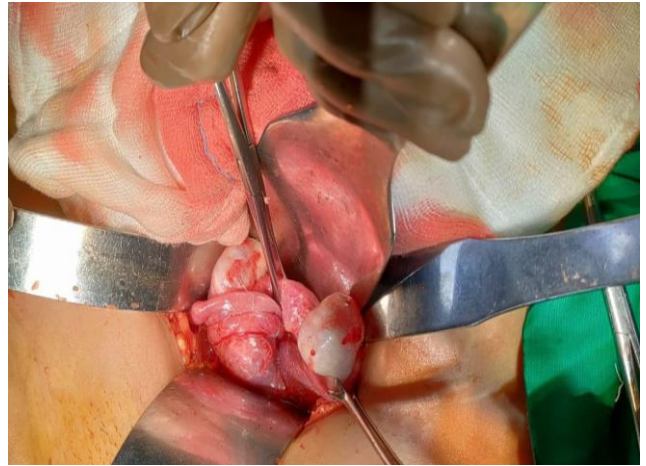
A 40 years unmarried nulliparous female denied sexual activity presented to Jagjivan Ram hospital (Western Railway), Mumbai at casualty on 08/02/2022 at 3:30 pm with complete uterine inversion. Patient was apparently alright till 4 hours back when suddenly noticed something coming out per vaginum while straining at defecation along with lower abdominal pain and minimal bleeding per vaginum. Patient also gave a history of one episode of nausea and vomiting 2 hours back. Patient had last menstrual period on 16/01/2022 with past menstrual cycles unremarkable and her urine pregnancy test was negative.



**Figure 1: Ultrasonography with filled urinary bladder and absent uterus in normal anatomical position.**



**Figure 2: A 10x9x8 cm fibroid with areas of congestion and degeneration with complete uterine inversion.**



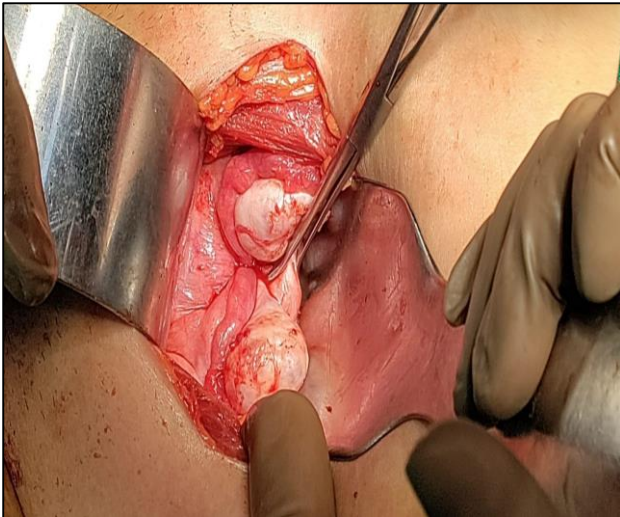
**Figure 3: Flower in vase appearance in complete uterine inversion.**



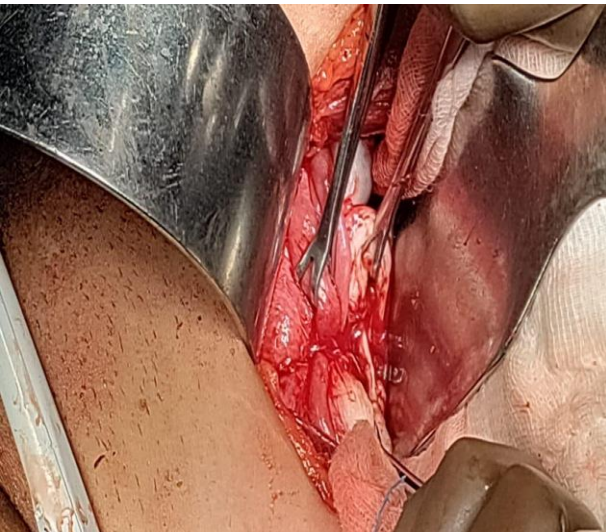
**Figure 4: Identification of plane between fibroid and fundus of the uterus.**



**Figure 5: Vaginal myomectomy.**



**Figure 6: Attempt to reposit the uterus by Huntington's procedure.**



**Figure 7: Attempt to reposit uterus by Haultain's procedure.**



**Figure 8: Reposition of uterus in anatomical position.**

Patient had a past history of menorrhagia 2 years ago for which USG was done suggestive of multiple uterine fibroids. Patient was started on tab mifepristone 25 mg once a day. Patient lost to follow up because of COVID pandemic and personal reasons. She had been consuming tabs for past 1 year and stopped medication 15 days ago on her own. Patient also had a history of chronic constipation. She had no history of urinary complaints, giddiness, syncope, palpitations, difficulty in breathing, foul smelling per vaginum discharge, fever, trauma, fall or local injury. On general examination she was conscious, cooperative, and well oriented with averagely built and nourished. She had mild pallor and she was vitally stable. Her respiratory and cardiovascular systems were within normal limits. Her abdomen was soft and no tenderness, no guarding, no rigidity with no palpable mass/ organomegaly.

On ultrasonography, a filled urinary bladder was visible and the uterus was not visualized in normal anatomical position. Bilateral ovaries were normal. There was no evidence of hydroureteronephrosis (Figure 1).

All routine investigations were within normal limits. After obtaining written, informed and valid consent, patient examined in dorsolithotomy position under general anesthesia in the operation theatre. External genitalia were normal. A firm red fleshy congested mass measuring approximately 10×9×8 cm seen coming out of introitus separating vulva with bosselated irregular surface and irregular margins. External OS of cervix could not be traced. No evidence of cystocele, enterocele and rectocele. On per rectal exam, uterus could not be felt at usual position and rectal mucosa was intact (Figure 2). Piece of necrotic tissue overlying mass removed and sent for HPE which reported as endometrium. Gentle and careful attempt of reducing the prolapsed mass was done however it could not be repositioned completely. It was deposited into vagina and packing was done with tampon soaked with betadine solution. Patient was vitally stable and tolerated procedure well. She started on injectable broad-spectrum antibiotics and was monitored in surgical ICU. Clinical and sonographic findings were suggestive of complete uterine inversion secondary to degenerated submucous fibroid polyp.

Patient and relatives were counselled in detail about present condition, possible treatment options and possible consequences and written, informed and valid consent was taken. She was posted for exploration via combined abdominal and vaginal approach SOS myomectomy SOS hysterectomy under spinal anesthesia after preanesthetic fitness. Under spinal anesthesia, in semi lithotomy position, there approx. 10×9×8 cm mass (fibroid) seen outside vagina. As tissue was oedematous and of the same consistency; it was not possible to identify the landmarks and structures to avoid any injury to pelvic structures. On opening abdomen via suprapubic transverse incision, an empty space was noted at site of the fundus of uterus along with approximation of both ovaries and fallopian tubes disappearing in constriction cup giving appearance of

flower in vase (Figure 3). After finding a plane between the fibroid and fundus of the uterus, vaginal myomectomy done after local infiltration of vasopressin. (Figure 4 and 5) With vaginal manipulation combined with abdominal traction to round ligament pedicle attempt of repositioning the uterus into abdominal cavity was made by Huntington procedure (Figure 6). After examining the anatomical position of urinary bladder and other pelvic structures, reposition of uterus in anatomical position done after incising posterior cervical constriction ring abdominally by Haultain procedure. (Figure 7 and 8). Total abdominal hysterectomy with bilateral salpingectomy with vaginal vault suspension was done sparing bilateral ovaries. Patient withstood the procedure well and was discharged on an uneventful postoperative 9<sup>th</sup> day.

Histopathology examination report confirmed diagnosis of leiomyoma uterus with areas of hemorrhage and degeneration with normal uterus and both fallopian tubes.

## DISCUSSION

Most of the uterine inversions present as obstetric complications. NPUI causes contribute to one sixth of all inversions.<sup>1,4</sup> They may be idiopathic or associated with uterine pathology like prolapsed fibroids, uterine neoplasm and endometrial polyp. Of these uterine leiomyomas contribute the maximum (78.8%-85%) number of cases.<sup>1,3</sup> The mechanism in which the tumours result in inversion of the uterus is largely unknown; however thin uterine wall, rapid growth of the tumour, dilation of the cervix by the tumour and its abrupt expulsion have been cited.<sup>1,3-5</sup> Fundal fibroids due to their traction effect have an increased risk.<sup>1,3,6</sup> Majority of non puerperal uterine inversion presents after 45 years and is mostly benign. Only 20% of pathologic specimens showed malignancy which were usually seen among young women.<sup>4,6</sup> Hence in women of reproductive age, malignancy is always a suspicion if uterine inversion occurs. This stresses on the need of histopathology specially to manage local recurrence or distant metastasis that may occur many years after the initial surgery.<sup>3,4</sup>

NPUI can pose a major diagnostic dilemma, since it is associated with non-specific clinical symptoms. Complaints such as lower abdominal pain, urinary disturbances, pressure, or a feeling of something coming down the vagina are often reported. In addition, patients may complain of vaginal discharge or vaginal bleeding.<sup>9</sup> These complaints are similar to other common gynecologic conditions, which may lead to misdiagnosis.

However clinical findings of a mass coming through the vagina (stage 3 or 4) without definite margins of a cervix and a nonpalpable uterus on bimanual or rectal examination can aid in the diagnosis.<sup>4,6</sup> The diagnosis of inversion may be difficult to make during examination especially with sloughed endometrium when it may be confused with fibroid polyp. The misdiagnosis of submucous fibroid and surgical attempt to remove it

vaginally may result in profuse bleeding and fundal perforation of the uterus.<sup>6</sup>

Uterine inversions can be classified as follows: Stage 1 Inversion of the uterus is intrauterine or incomplete. The fundus remains within the uterine cavity. Stage 2 A complete inversion of the uterine fundus through the fibromuscular ring of the cervix. Stage 3 Total inversion, whereby the fundus protrudes through the vulva. Stage 4 The vagina is also involved with complete its inversion through the vulva along with the inverted uterus.<sup>1</sup> Inversion can also be classified as acute and chronic.

Radiological investigations like ultrasound, computer tomography or magnetic resonance imaging can be used in stable patients. Ultrasonography (USS) should be the first line of investigation considering availability and simplicity. USS can help with both the diagnosis of NPUI and diagnosis of its aetiology. Sonographic characteristics of "Y"-shaped uterine cavity, in the longitudinal plane are seen in incomplete uterine inversions. The base of "Y" is the non-inverted endometrial lining. In contrast to incomplete inversion, the longitudinal view in complete inversion shows a "U"-shaped configuration, with the limbs of the "U" representing the complete inverted endometrial lining extending both anteriorly and posteriorly.<sup>10</sup> The 3D power Doppler with USS has been used more recently in the diagnosis of NPUI as it can clearly show the changes in the uterine artery course in relation to the uterine body. Zohov et al emphasized that once the vaginal probe (with 3D power Doppler) is applied directly to the uterine corpus of inverted uterus, it showed bilateral uterine arteries in a longitudinal central location along the uterine body, with a U-turn sign, showing a central course of the main uterine vessels instead of their normal anatomical peripheral location laterally alongside the corpus of the uterus.<sup>11</sup> MRI is found to be sensitive in the diagnosis of NPUI. The distinct observations identified are U-shaped uterine cavity, a thickened and inverted uterine fundus on a sagittal section, and a "bull's eye" configuration on the horizontal section.<sup>3,12-14</sup> This, however, is not readily available in most hospital settings of developing countries. CT scan has not been very useful in the diagnosis of NPUI. It can be an option in situations where MRI is not possible. The contrast-enhanced examination is favoured for delineation.<sup>15</sup>

In most of the case reports we reviewed, the difficulties of clinical diagnosis and interpretation of ultrasonography have been emphasized. To overcome this, examination under anaesthesia by experienced surgeons and histological sampling of the vaginal mass have been suggested. Demonstrating the endometrium on the surface of the mass will be confirmatory of the diagnosis.<sup>16</sup> Viewing the pelvis at laparoscopy or laparotomy is an alternative way to confirm NPUI if the imaging modalities fail to provide a reasonable diagnosis. The appearance of ovaries and tubes projecting out of the indented uterine fundus has been described as the "flower vase appearance" in cases of NPUI.<sup>17</sup>

Initial assessment and resuscitation would be the priority as some patients may be in septic or in hemorrhagic shock, followed by correction of anaemia, pain relief, and starting antibiotics. Once stabilized, all steps should be followed to confirm the diagnosis and to establish the possible aetiology. Unlike the timely reposition procedures which are successful in acute inversion; surgery is imperative in chronic cases. The type and approach of surgery should be individualized considering the age, desire for future fertility, aetiology (benign or malignant), and the stage of the disease in case of the malignancy. Surgery is the mainstay of treatment of NPUI, focusing on the repositioning of the uterus. Accordingly, surgical reposition or hysterectomy should be considered.<sup>3,6</sup> Repositioning is essential if uterine preservation is considered, as it is the only way to prevent pain, bleeding, infections, and gangrene. Hysterectomy is challenging if the uterus is not repositioned priorly because of the distortion of the pelvic anatomy in the inverted uterus as the ureters come in close proximity to the uterine vessels.<sup>4</sup> It should be assumed that hysterectomy would be technically easier on a normally positioned uterus, rather than the inverted uterus, as repositioning would restore normal anatomy with which gynecologists are familiar with. While stage 1 inversion will often offer easy repositioning of the fundus, inversions of stages 2, 3, and 4 are likely to be more demanding. Exclusion of malignancy and excision of the causative benign tumour are essential before repositioning and repair. If repositioning is impossible, the only option left would be hysterectomy. Authors believe that attempting to reposition a uterus with a malignancy would be detrimental as the peritoneal cavity would be exposed to the pathology through the incised uterine wall.

We recognized three main aspects to consider: (1) route of surgical access either abdominal (laparotomy or laparoscopy), vaginal, or both, (2) attempt to reposition, and (3) planning of the eventual surgical procedure (resection of the causative tumour and repair or hysterectomy). At times, a combination of approaches may be necessary to rectify this disorder.<sup>4,6</sup> There are 2 procedures by vaginal approach: Spinelli procedure is anterior and requires dissection of the bladder and an anterior uterine wall incision, while Kustner procedure is a posterior approach with incision on the posterior uterine wall which makes it easier and safer.<sup>1,4,6</sup> There are 2 procedures by abdominal approach: Huntington procedure 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 whereas the Haultain procedure uses a vertical incision in the posterior portion of the ring and gentle traction on the round ligaments.<sup>1,4</sup> These defects are then closed in layers in cases of uterine preservation; otherwise, hysterectomy is performed. Hysterectomy can be attempted without myomectomy to prevent excessive blood loss. But to aid in easy reposition of the uterus, one can first proceed with myomectomy. Most surgeons use the abdominal route for hysterectomy. However vaginal hysterectomy can be done even without reinverting.<sup>3,6</sup> Few studies have reported

nonpuerperal uterine inversion managed by laparoscopy assisted vaginal hysterectomy.<sup>4,6</sup>

There are reports of successful pregnancies following the surgical correction of puerperal uterine inversion. Surprisingly, even though the literature repeatedly says to conserve the uterus if fertility is required, we could not find any evidence of successful pregnancy following repositioning of a nonpuerperal uterine inversion. Irani et al. reported a case where the uterus was repositioned with the Haultain procedure, yet the woman remained subfertile for two years after the operation.<sup>18</sup> Should uterine preservation be successful, there is no evidence to suggest the appropriate interval before attempting pregnancies following these techniques; however, in other recommendations of pregnancy interval following uterine surgery, greater than 12 months is suggested.<sup>19</sup> As with all uterine surgery, the risk of uterine rupture with subsequent delivery should be addressed.

In our case, the patient developed acute inversion of uterus following straining at defecation with fibroid being the leading point of inversion of uterus. Unlike the case reported by Younas et al where the patient had hypovolemic shock and profuse bleeding, our patient was in a stable condition with lower abdominal pain and minimal bleeding per vaginum.<sup>6</sup> We confirmed the diagnosis with clinical examination and radiological investigations. After detailed counselling of patient and relatives, we proceeded with a combined abdominal and vaginal approach for further management. Following the vaginal myomectomy, an attempt was made to reposition the uterus by Huntington procedure which failed; after which the uterus was successfully repositioned by Haultain procedure. As our patient was not desirous of future fertility, we performed total abdominal hysterectomy with bilateral salpingectomy with vaginal vault suspension sparing bilateral ovaries. Shambhavi et al and De Vries et al also reported in their cases that the myoma was excised vaginally but hysterectomy was done abdominally.<sup>2,5</sup> Mwinyoglee et al reported an NPUI which was treated with vaginal hysterectomy without repositioning the uterus.<sup>20</sup> Mayadeo and Tank, described a case of in-complete lateral inversion of the uterus, diagnosed at laparoscopy, and treated with vaginal hysterectomy without repositioning the fundus.<sup>21</sup> Laparoscopic repositioning and repair were reported by Zhang et al. after releasing the anterior cervical ring during laparoscopy. They performed a vaginal myomectomy before repositioning.<sup>22</sup>

## CONCLUSION

With the rarity of this clinical condition, it is believed that most gynecologic surgeons will not encounter a case of NPUI during their careers.

Diagnosis requires a high index of suspicion and their management is a challenge to gynaecologists due to its rare occurrence, distorted pelvic anatomy and associated pelvic

organ injuries during surgery. Good anatomical and clinical knowledge along with surgical skills is of utmost importance for successful outcomes.

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