

## Non-Puerperal Uterine Inversion in a Nulliparous Woman with no Uterine Mass: A Case Report and a Literature Review

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### Summary

**Background:** Non-puerperal uterine inversion (NPUI) is an extremely rare condition. Most reported cases of uterine inversion were in multiparous women. Moreover, non-puerperal uterine inversion usually occurs when there is a benign or malignant uterine mass present. In literature, there have been only 9 reported cases of non-puerperal uterine inversion in a nulliparous woman in the 10 year window from 2006 to 2017 [1].

**Case:** Here, a case of non-puerperal uterine inversion is discussed. Initially the patient was diagnosed as a sub-mucosal fibroid of the uterus. The diagnosis of Non-puerperal uterine inversion was made only during operation. But the case reported in this paper, is the first reported case in a patient who did not have any uterine mass and also was nulliparous. At first, an attempt to reposition the uterus was made by Huntington method but was failed. Then, it was decided to perform the total abdominal hysterectomy. Her postoperative period was uneventful and she was discharged without complication after only seven days.

**Conclusion:** Non-puerperal uterine inversion is rarely encountered by Gynecologist. However, the rare occurrence of this case is often difficult to diagnose, especially when the exact cause of the condition is not known preoperatively. Our patient was lean, thin and malnourished, suffered from general weakness for long time. Could the weakness of the uterine muscle and ligaments be the cause for inversion? The reported case provides an indication for future research on the causes of non-puerperal uterine inversion, specifically the scenario which has no association to uterine mass in a nulliparous woman.

**Keywords:** Non-Puerperal Uterine Inversion, Sub-Mucosal Fibroid, No Uterine Mass, Nulliparous Woman.

### Introduction

Inversion of the uterus is a condition in which the uterus turns inside-out and the fundus prolapses to or through the cervix. Inversion of the uterus was classified by Jones into two types: puerperal or obstetric and non-puerperal or gynecologic [2]. As almost all cases of uterine inversion are puerperal, non-puerperal inversion of the uterus is an extremely unusual gynecologic condition. Uterine inversion can be described as follows as of first, second, third and fourth degree, shown in Figure 1.

- Incomplete inversion or 1st degree: The top of the uterus (fundus) has collapsed but the fundus doesn't come through the cervix.
- Complete inversion or 2nd degree: The uterus is inside out and fundus coming out through the cervix.
- Prolapsed inversion or 3rd degree: The fundus of the uterus is coming out of the vagina.
- Total inversion or 4th degree: Both of the uterus and vagina protrude inside out.

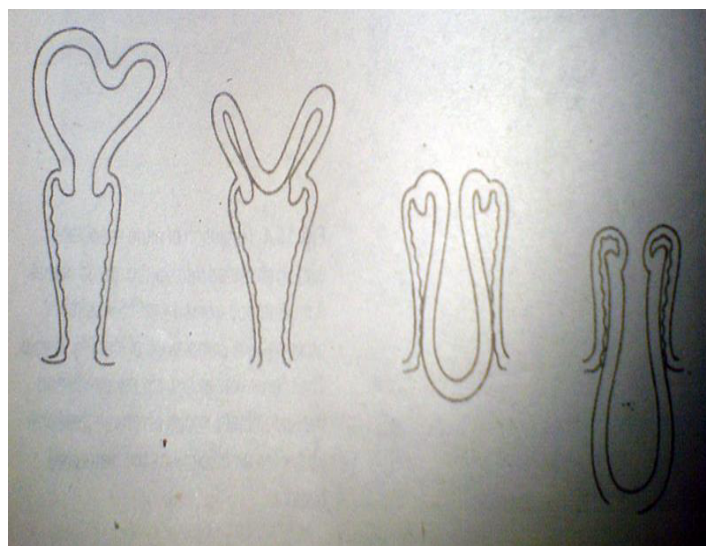


Figure 1: The degree of Uterine Inversion

There have been several researches in literature that shed light into particular features of non-puerperal uterine inversion. Most cases of non-puerperal uterine inversion occur in women over the age of 40 (Philippe Merviel et al.). Gomez-Lobo et al. analyzed 40 cases of non-puerperal uterine inversion between 1897 and 2018. The woman's age ranged from 15 to 88; 13 were primi-parous, 25 were multiparous, and 2 had never had sexual intercourse. Among them, only 4 cases were associated with women younger than 40. Range of the lesion size was from 2.9 cm (the teratoma) to 18 cm. The inversion was caused by malignancy in 65% of cases (26 cases), including sarcoma (leiomyo-sarcoma, rhabdomyo-sarcoma, and fibro-sarcoma [20 cases]), mixed Müllerian tumors (5 cases), and cervical adenocarcinoma (1 case). 13 cases had one or more myomas of the uterus and 1 case of teratoma in a 15-year-old patient. Mean age of the women was 44.3 for sarcoma, 46.9 for myoma, and 30.5 for the other lesions.

In their review study, Bruno Rosa Silva et al. presented 170 cases of Non-puerperal Uterine Inversion in the timeline from January 1940 to March 2017. The most common causes reported were leiomyoma (57.2%), sarcoma (13.5%), mixed mullerian tumor (4.7%), endometrial carcinoma (4.7%), Idiopathic (9.9%), Carcinoma (3.5%), rhabdomyo-sarcoma (2.9%), endometrial polyp (1.8%), chronic elevated intra-abdominal pressure due to liver cirrhosis and ascites (1.2%) and teratoma (0.6%).

Literature has only two cases of non-puerperal uterine inversion reported so far that were without any underlying pathology: one was in a postmenopausal lady of 67 years old, para 8+0, all by normal vaginal deliveries. Another one was in a 35 year old grand multipara women P8+0 (5 alive), all by spontaneous vaginal deliveries.

However, the case presented in current study was in a nulliparous lady with no indications mentioned above what so ever. However, this patient was lean, thin, and malnourished throughout her whole life. She also suffered from general weakness for a long period which might triggered the starting process of the uterine inversion.

Non-puerperal uterine inversions are rare cases with limited literatures on its incidence and causes [3, 4]. Non-puerperal inversions are commonly caused by benign sub-mucosal fibroid while others include sarcoma, malignant mixed mullerian tumor and endometrial polyp [5-8].

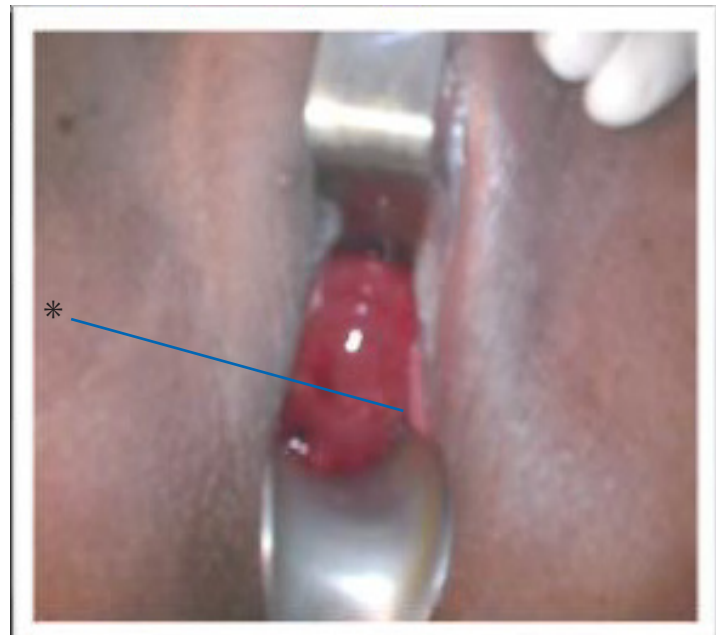
This article reports an extremely rare case of non-puerperal uterine inversion in a nulliparous woman without any benign or malignant uterine tumor. It was very difficult case to diagnosis preoperatively and is the first reported case of a non-Puerperal Uterine Inversion without any uterine mass in a nulliparous woman.

### Case Report

A 40-year-old nulliparous woman was admitted to the Ganashasthaya Shamajvittik Medical College & Hospital with complaints of irregular per vaginal bleeding and occasional lower abdominal pain for five years, which had increased with fever for last four days. The patient never conceived and suffers weakness for a long period. The patient's age of menarche was at 11 years old and her menstrual cycle was regular accept for the last 5 years. Her regular menstrual period was 4-5 days but during last 5 years, it stayed up to 10-12 days. The patient was lean and thin weighing

45 kg. Her temperature was 100°F and had no hypertension or diabetic. The history revealed that she had no sexual difficulties for the first 3-5 years of 20 years of marriage. Last 15 years, her sexual life had difficulties with occasional bleeding after sex and discomforts. She never sought any medical attention for it.

The abdominal examination showed soft abdomen, slightly tender on supra-pubic region and no mass was palpable in the lower abdomen. The speculum examination found a big mass in vagina, reddish in color, 6 cm x 5 cm in size, bled on touch and pus like discharge was seen as shown in Figure 2. The bimanual per vaginal examination showed that cervix was not properly palpable with palpable mass; firm in consistency (6 cm x 5 cm in size), bled on touch; and uterus was not palpable properly (rush due to bleeding and tenderness).



**Figure 2:** Closer view of the per speculum examination. Red color mass like structure formed in vagina.

These evidences give signals for a sub-mucosal fibroid. Therefore, the patient was clinically diagnosed, as a sub-mucosal fibroid of the uterus with pelvic inflammatory disease (PID) might be endometritis.

The ultra-sonogram (USG) reports of lower abdomen are shown in Figure 3:

- Urinary Bladder- normally filled and no intra vesicular lesion is seen;
- Uterus-bulky with a fibroid, lower part of the uterus measuring 40mm x 43mm;
- Tube-ovarian regions are not clearly seen and no pelvic collection is seen;
- Comment-Fibroid Uterus.



**Figure 3:** Shows the USG image

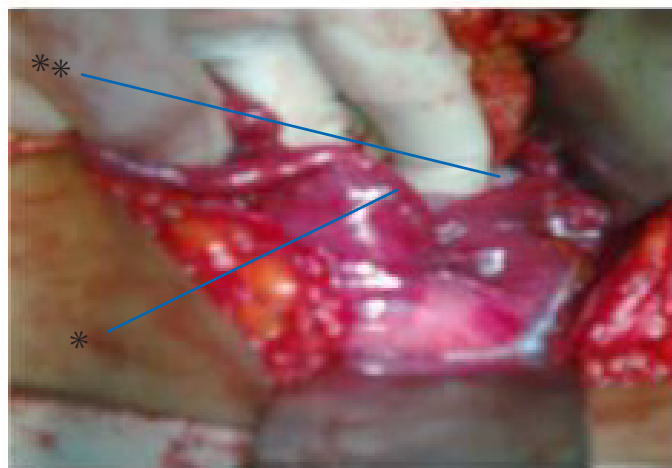


**Figure 4 (b):** View at laparotomy

The other investigation reports are:

- Complete blood count: -RBC: 4.5/cu mm -Hb: 11gm/dl -WBC: 7440/cu mm -ESR: 25 mm in 1st hour -Platelet: 155000/cu mm -DC: Normal limit;
- Blood group: B+ (positive) VDRL: Nonreactive RBS: 3.4 mmol/L HBsAg: Negative;
- Urine R/M/E, Echocardiogram and chest X-Ray were within normal limit.

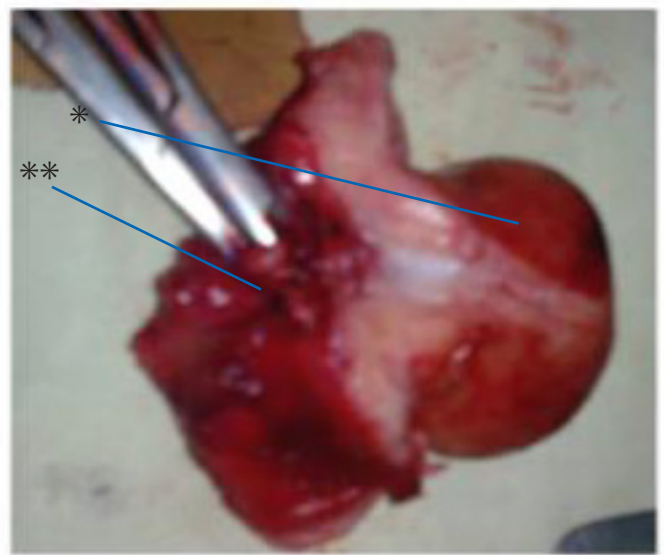
Since the patient was admitted to the hospital, she was treated with antibiotics such as Ciprofloxacin for 7 days and Metronidazole for 5 days. After 7 days of admission, when her temperature became normal and lower abdominal pain was subsided, the patient was taken for operation under spinal anaesthesia. During the laparotomy, uterus looked abnormal, both ovaries and fallopian tubes (distal part) found to be in the pelvis but outside of the concavity of the inverted uterus as shown in Figure 4(a) and 4(b).



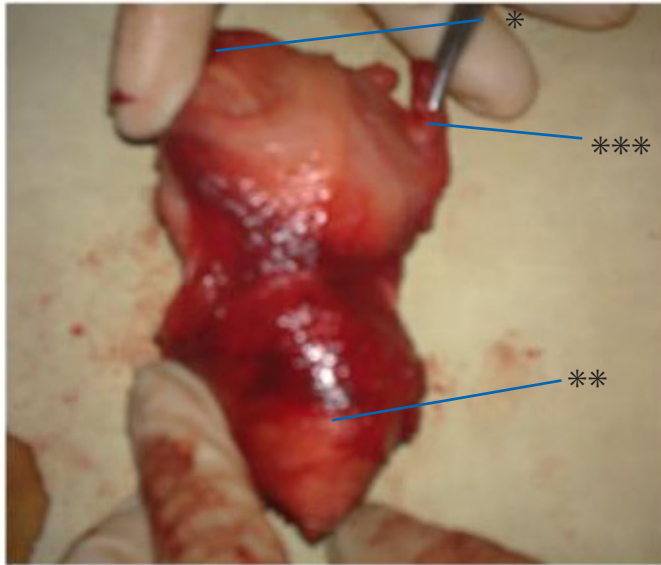
**Figure 4 (a):** View at laparotomy. The fallopian tubes (\*) are drawn inside the inverted uterus but the ovaries (\*\*) are clear of the constricting ring.

There was a small cyst (1x0.5 cm) in the right ovary, which was punctured. The diagnosis of **uterine inversion** was only made then, **during operation**. As there was no previous history that can be indicative of a possible inversion of the uterus, this diagnosis was very much unexpected, surprising and thus naturally quite nerve wrecking for the surgeon. At first, an attempt to reposition the uterus was made by Huntington method but was failed. Then, it was decided to perform the total abdominal hysterectomy (TAH). This procedure was successfully done although took longer (about two hours) than my usual TAH (performed in about 40-50 minutes). There was blood loss of about 150 ml.

Figure 5(a) shows the post-operative view of the inverted uterus, which clearly reveals non-puerperal uterine inversion of the uterus. Moreover, Figure 5(b) shows post-operative view of corrected uterus and cervix, which shows the normal uterus in size and shape without any benign or malignant uterine mass. Histopathology report also confirmed the absence of uterine mass of any kind.



**Figure 5(a):** Post-operative view of inverted uterus showing the inverted fundus (\*) and part of the fallopian tube (\*\*)



**Figure 5(b)** Post-operative view of corrected uterus and cervix which are normal in size and shape, leaving no trace of fibroids or any other abnormality. The fundus (\*), Cervix (\*\*) and intramural part of the fallopian tube (\*\*\*), is clearly visible.

After operation, specimen was sent for histopathology and the reports are as follows:

- **Gross:** Specimen consists of resected uterus without adnexa measuring 8x4x3 cm. Three blocks were embedded.
- **Microscopy:** Sections of the uterine wall showed unremarkable myometrium and the endometrium was thin with scanty gland and infiltrations of lymphocytes and plasma cells in the stroma. Sections of the cervix showed features of chronic cervicitis with squamous metaplasia in the lining epithelium. No evidence of malignancy was seen.

**Diagnosis:** Uterus (Resected), chronic endometritis. Chronic cervicitis with squamous metaplasia.

#### Outcome & Follow Up:

The patient's post-operative period was uneventful and was discharged on the 7th postoperative day without any complications. First follow up was done after 15 days of discharged before going back to her village and she had no complaints. The second follow up was made after two months of the first follow up and the patient was fine and felt relief from any discomforts or difficulties. It should be noted that there was proper counselling during discharge and every follow up meetings.

#### Discussion

Most commonly, puerperal uterine inversion arises as a complication of delivery [9]. The most common type called acute inversion is defined as when inversion occurs during the first twenty-four postpartum hours. While sub-acute inversion occurs after twenty-four hours but within four weeks of delivery. Sometimes it occurs after four weeks of delivery and is termed as chronic inversion of the uterus. Non-puerperal uterine inversion is often of the chronic type. It is also reported that 8.6% of non-puerperal uterine inversion are sudden onset [10].

Incidence of puerperal uterine inversion is approximately 1 in 30,000 to 1 in several hundred thousand labors [2, 11, 12]. But Non-puerperal uterine inversion is a rarer condition occurring in approximately 5 - 17 % of all uterine inversions [13, 14]. There has been several studies reviewing the cases of uterine inversion in various timeframes. In almost all of them, the number never exceeded beyond hundreds for the last century.

A review study done in the timeframe from 1940 to 2000 by Takano et al. revealed 88 cases of non-puerperal uterine inversion. Among which 92% was associated with tumors. Among those, 71.6% was leiomyoma, 20% was malignant tumor (endometrial carcinoma 6.8% and sarcoma 13.6%) and the rest (8%) was idiopathic [12].

On the other hand, Bruno Rosa Silva et al. reviewed papers published in the literature from 1940 up until March 2017 to recover a total of 229 cases reported. They performed a systematic review about non-puerperal uterine inversion that included 170 case reports published demonstrating the rarity of this pathology.

Magnetic resonance imaging (MRI) is a useful diagnostic equipment for proper diagnosis of non-puerperal uterine inversion to examine the characteristic image of uterine inversion. A 'U' shaped uterine cavity, a thickened and inverted uterine fundus on a sagittal image and a "bull's-eye" configuration on an axial image indicative of uterine inversion [15]. Ultrasonography examination is also a useful diagnostic tool. Chou et al reported that an indentation of the fundal area was observed on a longitudinal scan of the uterus by trans-abdominal ultrasonography [16]. Hsieh et al reported that a longitudinal abdominal ultra-sound scan revealed a depressed longitudinal groove extending from the fundus to the center of the inverted portion [17].

Non-puerperal uterine inversion is a very unusual condition that most of the gynecologists would never encounter and thus must be dealt with little or no previous experience. The etiology of the inversion of uterus is not clearly defined and can be multifactorial. The following etiologies have been described in the literature: rapid growth of the tumor, a fundal location of the tumor, a thin uterine wall, and dilatation of the cervix [18]. It has also been suggested that raise of intra-abdominal pressure during coughing, sneezing, and straining may be involved in the development of uterine inversion [19, 20].

An interesting feature of this reported case is that no obvious reason was found so as to why the uterus became inverted, as the patient is nulliparous with no fibroid, polyp or malignant tumor from uterus, which might have started the inversion process. But patient was lean and thin and suffered weakness for a long period. Can weakness of the uterine muscle and ligaments be a viable reason? Therefore, this reported case is an extremely rare one and the first one reported. The author never anticipated the inversion as the patient was nulliparous and did not have any tumor from uterine origin. Moreover, the examination findings (described above) and ultra-sonogram reports (fibroid uterus) were misleading and created confusion before operation. Thus, initially it was diagnosed as a sub-mucosal fibroid of the uterus.

There were no reports of MRI and CT scans as those tests are expensive and patient could not afford them. These were the reasons why pre-operative diagnosis of the case was difficult and

proper diagnosis of non-puerperal uterine inversion was made during operation.

At the time, the author found this to be the only case of non-puerperal uterine inversion of her 27 years of surgical life experiences. However, later in 2012, a fellow young colleague, upon facing difficulty during a caesarean section and unable to assess the scenario asked for the author's consult. Experienced by the case presented in the current study, the author readily identified the case in 2012 to be a uterine inversion. This second encounter of uterine inversion was however in a parous woman and thus was a puerperal uterine inversion. Sometimes, gynecologists may not encounter a case of uterine inversion during their lifetime.<sup>20</sup> Therefore, pre-operative diagnosis is essential for appropriate management.

### Conclusion

To conclude, this case of non-puerperal uterine inversion is an extremely rare one diagnosed at laparotomy with no descriptive cause discovered in a nulliparous woman. However, this rare case of non-puerperal uterine inversion is often difficult to diagnosis especially when the exact cause of this case is still unknown. The reported case provides an indication for future research on the causes of non-puerperal uterine inversion.

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### References

1. Alshabi J, Alsomairi A, Elmuzaini F (2019) Non-puerperal uterine inversion due to submucosal fibroid in a nulliparous woman: A case report. *Int J Case Rep Images* 10: 101008Z01JA2019.
2. Jones HW (1951) Non-puerperal inversion of uterus. *Am J Surg* 81: 492-495.
3. Kopal S, Seckin NC, Turhan NO (2001) Acute uterine inversion due to a growing sub-mucous myoma in an elderly woman: case report. *Eur J Obstet Gynecol Reprod Biol* 99: 118-120.
4. Pride GL, Shaffer RL (1977) Non-puerperal uterine inversion: Report of an unusual case. *Obstet Gynecol* 49: 361-364.
5. Sharma JB, Kumar S, Rahman SM, Roy KK, Malhotra N (2009) Non-puerperal incomplete uterine inversion due to large sub-mucous fundal fibroid found at hysterectomy: a report of two cases. *Archives of Gynecology and Obstetrics* 279: 565-567.
6. Buyukkurt S, Vardar MA, Zeren H, Ozgunen FT (2007) Non-puerperal inversion of the uterus caused by leiomyosarcoma: A case report and clinical management. *J Obstet Gynaecol Res* 33: 402-406.
7. Cormio G, Loizzi V, Nardelli C, Fattizzi N, Selvaggi L (2006) Non-puerperal uterine inversion due to uterine sarcoma. *Gynecologic and Obstetric Investigation* 61: 171-173.
8. Eftekhari Z, Rahimi-Moghaddam P, Izadi-Mood N, Yarandi F (2005) Non-puerperal uterine inversion caused by uterine sarcoma. *Australian & New Zealand Journal of Obstetrics & Gynaecology* 45: 82-83.
9. Beringer RM, Patteril M (2004) Puerperal uterine inversion and shock. *British Journal of Anaesthesia* 92: 439-441.
10. Das P (1940) Inversion of the uterus. *J Obstet gynecol Bp Emp* 47: 527-548.
11. Abouleski E, Ali V, Joumaa B, Lopez L, Gupta D (1995) Anaesthetic management of acute puerperal inversion. *Br J Anaesth* 75: 486-487.
12. Takano K, Ichikawa Y, Tsunoda H, Nishida M (2001) Uterine inversion caused by uterine sarcoma: a case report. *Jpn J Clin Oncol* 31: 39-42.
13. Dali M, Rajbhandari S, Shrestha S (1997) Puerperal inversion of the uterus in Nepal: case reports and review of literature. *J Obstet Gynaecol Res* 23: 319.
14. Fofie C, Baffoe P (2010) Non-puerperal uterine inversion: a case report. *Ghana Med J* 44: 79-81.
15. Lewin JS, Byyon PJ (1989) MR imaging of uterine inversion. *J Comput Assist Tomogr* 13: 357-359.
16. Chou HH, Chen CI, Chu KK (1995) Ultrasonography diagnosis in sub-acute uterine inversion-A case report. *Changcheng Yi Xue Za Zhi* 18: 73-76.
17. Hsich TT, Lee JD (1991) Sonographic findings in acute puerperal uterine inversion. *J Clin ultrasound* 19: 306-309.
18. Rosales Aujang E, Gonzales Romo R (2005) Nonpuerperal uterine inversion. *Gynecolobstet Mex* 73: 328-331.
19. Lupovitch A, England ER, Chen R (2005) Non-puerperal uterine inversion in association with uterine sarcoma: case report in a 26-year-old and review of the literature. *Gynecol Oncol* 97: 938-941.
20. Mwinyoglee J, Simelela N, Marivate M (1997) Non-puerperal uterine inversions: A two case report and review of literature. *Cent Afr J Med* 43: 268-271.

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