

## Case Report

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# Inside out: A successful repositioning of a chronic third-degree uterine inversion

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### Abstract:

In this day and age, with the advancement of treatments and the strong campaign to discourage home deliveries, chronic uterine inversion is now considered an uncommon but life-threatening obstetric condition. In this report, we present a case of a 17-year-old primipara, who sought consultation due to prolonged and heavy vaginal bleeding. Upon speculum examination, a knob-like, fleshy, hyperemic, smooth mass, approximately 6 cm × 5 cm × 4 cm, was seen occupying the upper third of the vaginal canal. On internal examination, a globular mass was felt protruding through the cervix, which bleeds easily on manipulation. Uterine corpus was neither appreciated on both abdominal and rectovaginal examination. The patient was admitted and managed as a case of chronic uterine inversion, which was further confirmed by a sonogram. Successful repositioning of the uterus was achieved after trying different established techniques and procedures in the attempts at maneuvering the chronically inverted uterus.

### Keywords:

Haultain, Huntington, Johnson maneuver, uterine inversion

## Introduction

Uterine inversion is a rare obstetric complication that has an incidence of 1 in 2,000–1 in 50,000 live births.<sup>[1]</sup> Nowadays, the reported incidence is declining because of improved health-seeking behavior and increasing institutional deliveries assisted by health professionals. However, in countries like the Philippines, where traditional birth attendants are still very much relied on because of cultural and financial reasons, home deliveries are still prevalent. This mainly contributes to the prevalence of mismanaged third stage of labor.

Puerperal uterine inversion is the introflexion of the uterine body protruding

into the vagina or out of the vulva.<sup>[2]</sup> It is more common than nonpuerperal uterine inversion, which is often seen in association with gynecologic pathology. Puerperal uterine inversion is classified as acute, subacute, and chronic, with the prevalence of 83.4%, 2.62%, and 13.9%, respectively.<sup>[3]</sup>

Due to the rare occurrence and limited literature on chronic uterine inversion, its diagnosis is often missed and is later on recognized at an advanced degree and severity. Early detection of uterine inversion is therefore vital in attaining good outcomes. Conversely, late diagnosis may result in massive bleeding and shock, eventually leading to death.

## Case Report

A 17-year-old G1P1(1001) sought a consult at the emergency room due to vaginal bleeding of 4 months duration.

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Eight months prior to consultation, the patient delivered to a full term, cephalic, livebirth, baby boy via spontaneous vaginal delivery at home attended by a traditional birth attendant.

One day prior to her delivery, the labor pain started at 1 to 2 hour interval. Labor pain progressed, prompting them to seek help from a known local traditional birth attendant. Internal examination revealed a 4-cm dilated cervix. Approximately 9 hours after, abdominal manipulation with the use of an herbal oil was allegedly administered for an hour and was repeated after 2 hours.

Six hours after the abdominal manipulation, the patient was asked to bear down. Bearing down was allegedly ineffective, and thus, fundal pressure was also applied by another person by pushing the fundal area of the uterus toward the vagina. After an hour and a half of bearing down and fundal pressure, a big baby with the birthweight of 4.2 kg was delivered and was observed to be bluish in color. The baby eventually cried and turned pink after continuous rubbing of the back. Fifteen minutes after delivery of the baby, there was spontaneous delivery of the placenta, which was described to be crushed in appearance. There was also a perineal laceration of which degree was unknown; however no episiorrhaphy was done. She was then instructed to take the following medications: tranexamic acid, ferrous sulfate, and multivitamins. No antibiotics or uterotonics were given.

There was persistence of postpartum bleeding for the next 3 days, consuming 2 fully-soaked adult diapers per day, associated with dizziness and headache. No medical consultations were done. Four days after delivery, the amount of bleeding decreased to just spotting and totally stopped 7 days after delivery. The aforementioned medications were taken for 1 week. Breastfeeding was attempted but failed because the patient claimed that there was no enough milk that can be expressed, and thus they resorted to bottle-feeding.

During the interim, the patient was apparently well, until four months prior to consultation, she noted heavy vaginal bleeding, consuming 2-3 fully-soaked baby diapers per day which lasted for 7 days. This was associated with dizziness and intermittent hypogastric pain. There were also noted dyspareunia and post-coital bleeding. She did not seek consult nor did she take any medications.

Two months prior to consult, increasing amount of vaginal bleeding was noted, now consuming 4-5 fully-soaked baby diapers per day, associated with dizziness and hypogastric pain.

Persistence of these signs and symptoms, now with

generalized body weakness and headache 4 days prior to consult, prompted consult at a local hospital. She was given hematinics and was advised blood transfusion, thus the need for transfer to our institution.

The patient has unremarkable prenatal, past medical, family, social, menstrual, and sexual history.

The patient came in at the emergency room conscious, coherent, normotensive, and tachycardic. She was weak-looking, with pale palpebral conjunctivae. Breath sounds were clear and equal. The abdomen was flat with normoactive bowel sounds, tympanitic on all quadrants, soft, nontender, with no palpable pelvoabdominal mass. External genitalia showed an approximately 1-cm scar at the left, posterior fourchette of the vagina [Figure 1]. Speculum examination showed a knob-like, fleshy, hyperemic, smooth mass, approximately 6 cm x 5 cm x 4 cm, bleeding and occupying the upper third of the vaginal canal [Figure 2]. No vaginal discharge was noted. On internal examination, a globular mass, measuring 6 cm x 5 cm x 4 cm, is felt protruding through the cervix, which bleeds easily on manipulation. Uterine corpus was neither appreciated on both abdominal and rectovaginal examinations. No adnexal masses were noted as well. Rectovaginal examination revealed a tight sphincteric tone. The posterior portion of the prolapsed mass could be palpated. She was subsequently admitted with the diagnosis of G1P1(1001), Abnormal uterine bleeding probably secondary to chronic uterine inversion, 3<sup>rd</sup> degree, versus prolapsed submucous myoma, uterine sarcoma, cervical myoma, or cervical carcinoma, anemia, severe.

On admission, laboratory examinations revealed a very low hemoglobin level at 39 g/L and hematocrit at 0.15. The management plan was for correction of the anemia and repositioning of the uterus. There was also contemplation of a total abdominal hysterectomy should attempts at repositioning fail. The surgical plans were thoroughly discussed with the patient and her partner, and corresponding consent was secured. She was



Figure 1: Examination of the external genitalia shows an approximately 1-cm scar at the left, posterior fourchette of the vagina

transfused with 4 units of packed RBC, which increased her hemoglobin level to 94 g/L and hematocrit level to 0.30.

On the 4<sup>th</sup> day of admission, transvaginal and pelvic ultrasound revealed the following findings: inverted uterus or upside down uterine fundus, occupying the vaginal vault in its sagittal view, measuring 4.52 cm x 4.90 cm x 4.38 cm, with a pseudo-stripe sign [Figure 3], representing the apposed uterine serosa. On transverse view, the uterine fundus was observed between the vaginal walls, giving a typical target sign [Figure 4]. These all confirmed the diagnosis of a uterine inversion. No abnormal mass or free fluid in the cul-de-sac were seen.

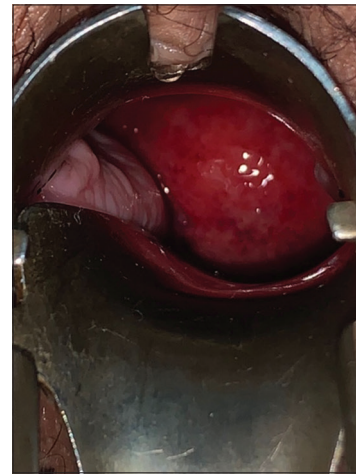
It was only on the 6<sup>th</sup> day of admission that a negative COVID 19 reverse transcription – polymerase chain reaction swab was released, allowing us to proceed to our contemplated procedures.

Attempts at manual repositioning of the uterus using Johnson's maneuver under generalized endotracheal anesthesia were unsuccessful. We then proceeded to attempting laparoscopic Huntington and Haultain procedures. Upon direct visualization of the intraabdominal organs, a flower-vase appearance with the round ligament, fallopian tubes, and ovaries projecting out of the indented uterine fundus was noted [Figure 5]. Huntington's procedure was attempted by grasping and pulling the round ligaments using Maryland grasper through the indented uterine fundus [Figure 6]. This was followed by the Haultain's procedure by making a vertical incision approximately around 2 to 3 cm on the posterior rim of the constriction ring [Figure 7]. There was only incomplete repositioning of the inverted uterus and further attempts proved futile due to the rigid constriction ring, thus the need to proceed to a laparotomy. Using a Babcock, the right and left round ligaments were pulled up with simultaneous pressure being applied on the inverted fundus through the vagina by an assistant. The initial incision on the constriction ring was extended toward the posterior lower uterine segment approximately 5 to 6 cm. Complete repositioning [Figure 8] was followed by repair of the vertical incision, which was done in layers [Figure 9]. The first layer was done through a continuous interlocking technique using Vicryl 1 atraumatic suture. The second layer was repaired by doing football stitches using Vicryl 1 atraumatic suture. Blood loss was minimal at approximately 400 cc. The patient tolerated the procedure well.

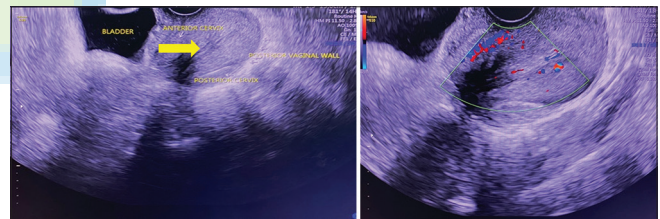
Post-operative course in the ward was unremarkable. Etonogestrel subdermal implant (Implanon) was administered prior to discharge. She was then advised the need for cesarean section for her succeeding

deliveries. The patient was discharged improved on postoperative day 3.

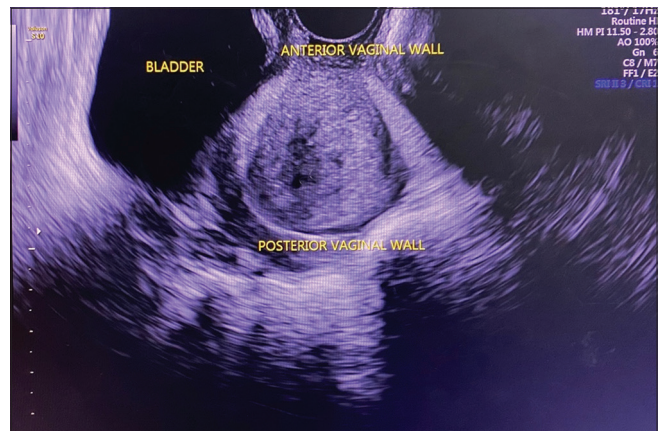
She came back for follow-up 4 months after the operation without any subjective complaints, fully recovered. She denied any abnormal vaginal bleeding, dyspareunia, coital bleeding, and hypogastric pain. Repeat transvaginal ultrasound revealed a normal-sized



**Figure 2:** Speculum examination: Vaginal canal is smooth and pink. A knob-like, fleshy, hyperemic, smooth mass, approximately 6 cm x 5 cm x 4 cm, was seen bleeding and occupying the upper third of the vaginal canal. No vaginal discharge was noted



**Figure 3:** Sagittal view revealed pseudo-stripe sign (yellow arrow, left picture), representing the apposed uterine serosa with inverted uterine vessels (right picture) spanning the entire uterine length



**Figure 4:** Transabdominal. On transverse view, the uterine fundus was observed between the vaginal walls, giving a typical target sign



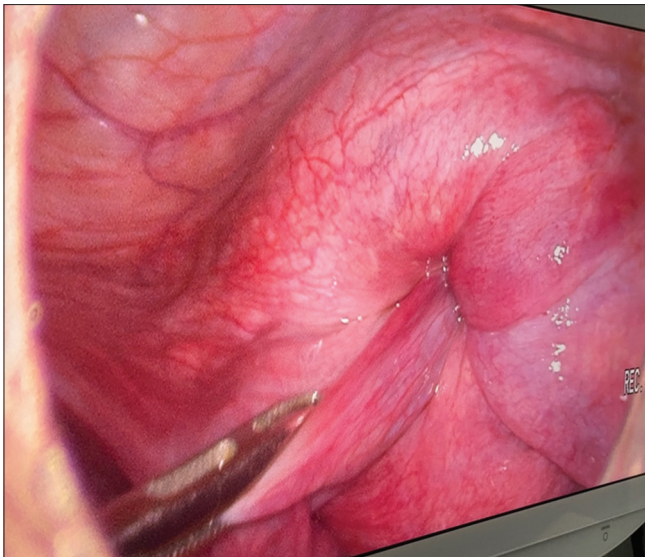


Figure 5: Flower-vase appearance with the round ligament, Fallopian tubes, and ovaries projecting out of the inverted fundus

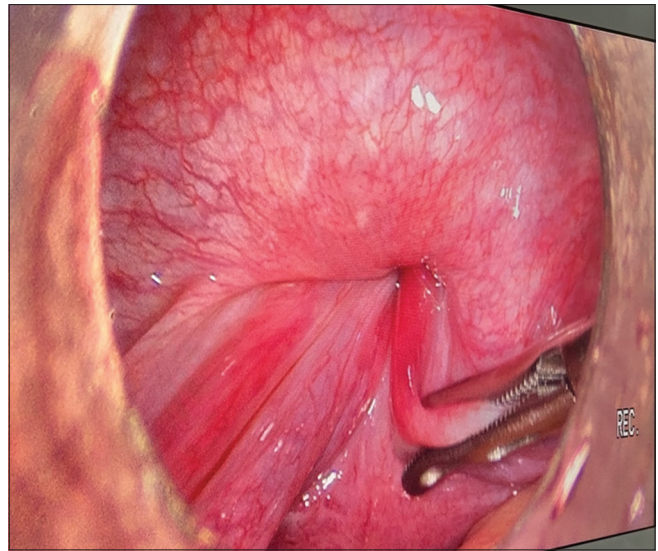


Figure 6: Huntington's Procedure: Grasping of the round ligaments and the uterus below the area of inversion

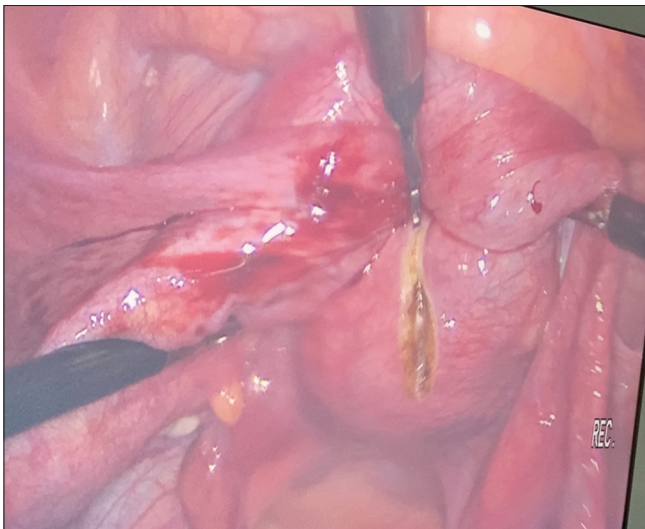


Figure 7: Haultain procedure: Incising the posterior part of the constriction ring

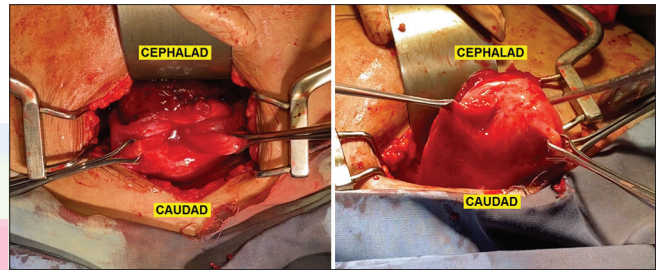


Figure 8: Eversion of the inverted uterus

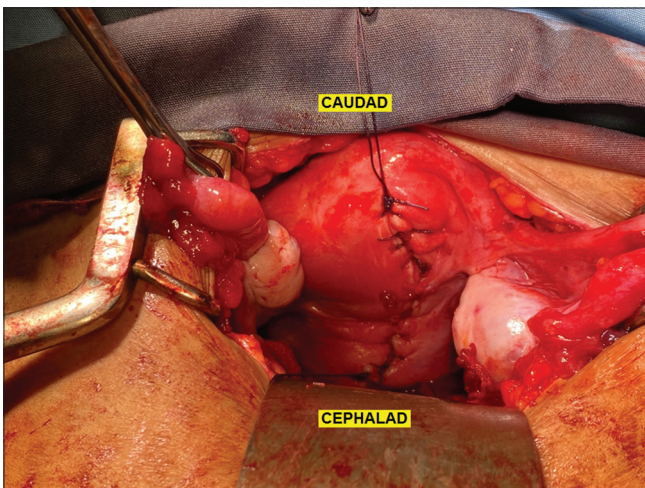


Figure 9: Repair of the vertical incision on the posterior part of the uterus

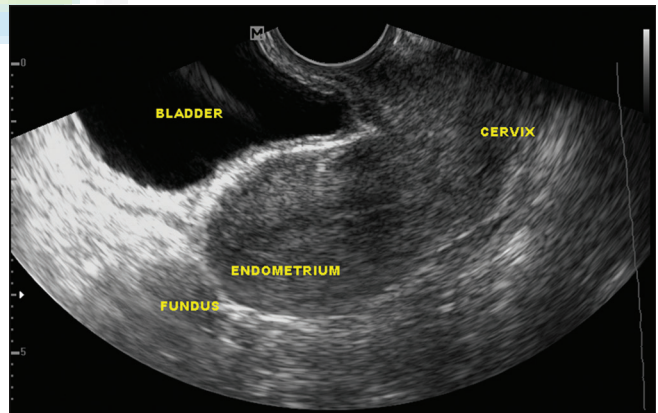


Figure 10: Repeat transvaginal ultrasound 4 months post operation reveals a normal-sized anteverted uterus with mild posterior wall adenomyosis, and proliferative phase-like endometrium

anteverted uterus with mild posterior wall adenomyosis, proliferative phase-like endometrium, normal ovaries, and no free fluid in the cul-de-sac [Figure 10].

### Case Discussion

Uterine inversion is defined as the introflexion of the uterine body protruding into the vagina or out

of the vulva.<sup>[2]</sup> It is a rare, life-threatening obstetrical complication of a mismanaged third stage of labor. It can either be acute or chronic in nature. If not recognized and treated promptly, it can result in massive bleeding and shock, which may eventually lead to maternal death.<sup>[4]</sup>

The reported incidence of uterine inversion ranges from 1 in 2,000 to 1 in 20,000 vaginal deliveries.<sup>[5]</sup> In our institution, there were six reported cases of acute uterine inversion for the past 5 years, from 2015 to 2019. All six underwent successful manual repositioning of the uterus, using Johnson's maneuver. This is the first documented case of a chronic uterine inversion in our institution.

Uterine inversion happens when a degree of uterine relaxation is combined with a simultaneous downward movement of the fundus such as during aggressive fundal pressure or excessive traction on the umbilical cord, followed by the resumption of uterine contractions.<sup>[6]</sup> The exact cause of uterine inversion may not be clear, but multiple predisposing factors are known such as fundal placental implantation, uterine atony, accrete syndromes, short umbilical cord, fetal macrosomia, primiparity, excessive fundal pressure, cord traction applied before placental separation, and rapid emptying of the uterus after prolonged distention.<sup>[5,7,8]</sup> The latter five risk factors were present in our patient.

The process of uterine inversion usually starts in the fundus with variable degrees and severity of inversion. Uterine inversion can be classified anatomically or chronologically.<sup>[9]</sup> Anatomical classification is as follows [Figure 11]: first degree, when a portion of the uterine fundus indents toward the endometrial cavity or mere dimpling of the fundus; second degree, the inverted fundus extends to but not through the cervix; third degree, the inverted fundus extends through

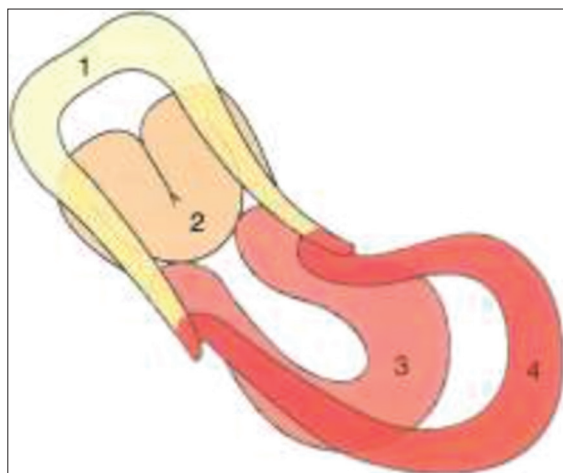


Figure 11: Progressive degrees of uterine inversion

the cervix but remains within the vagina; and fourth degree, the inverted fundus extends outside the vagina.<sup>[5]</sup> Chronological classification is based on its diagnosis from the time of delivery. It is classified as acute, occurring within 24 h of delivery; subacute, occurring 24 h to 30 days postpartum; and chronic, occurring more than 30 days postpartum.<sup>[10]</sup> Our index patient was classified as chronic uterine inversion, third degree.

Uterine inversion is diagnosed clinically. Its clinical presentation may vary with various degrees of inversion, from mere pain to hemorrhagic shock. The diagnosis of uterine inversion is often missed at the time of delivery, especially those of the first and second degree due to mild or absent symptoms. This may later progress to chronic inversion to the third or fourth degree, as seen in our patient.

Clinical manifestations usually include abnormal uterine bleeding, foul-smelling vaginal discharge, or a mass protruding through the cervix.<sup>[1]</sup> Other accompanying symptoms can be chronic pelvic pain, low back pain, and history of postpartum hemorrhage.<sup>[3]</sup> The most common sign encountered is the observation of the uterine fundus beyond the vaginal introitus or palpation of the fundus through the external os. In our case, a globular mass was felt and seen protruding through the cervix to the upper third of the vaginal canal.<sup>[10]</sup>

When the physical examination is equivocal and the patient is hemodynamically stable, sonographic evaluation is of value in clinching the diagnosis of a uterine inversion.<sup>[10]</sup> There are distinct sonographic characteristics of an inverted uterus. First would be a sonographic finding of a mirror reflection of the normally positioned uterus projecting into the vagina. Second would be a pseudostripe, which is formed by the interface of the two opposing serosal surfaces that mimic an endometrial stripe [Figure 3]. Third would be the target sign, created by the uterine fundus, lined by a hyperechoic endometrium, surrounded by a hypoechoic rim, representing fluid within the space between the inverted fundus and the vaginal wall [Figure 4].<sup>[9]</sup> The aforementioned sonographic characteristics were all seen in our patient.

In a chronically inverted uterus, such as in our case, the involution of the cervix induces a constriction ring, making the restoration of the normal position of the uterus difficult.<sup>[7]</sup> The goal is to divide the constriction ring either anteriorly or posteriorly.<sup>[1]</sup> Once it is identified surrounding the inverted dome of the uterus, it is incised vertically at its posterior or anterior rim. This increases the ring size and enables repositioning of the uterus.<sup>[7]</sup> A delay in the diagnosis of uterine inversion will require a more invasive surgical intervention due to the increasing



firmness of the constriction ring. The chronically inverted uterine walls have reduced resilience resulting from the complete involution.<sup>[1]</sup> Surgical approach is also indicated when attempts at manual reduction fail.

There are several surgical approaches for repositioning uterine inversion. The vaginal route is comprised Spinelli and Kustner techniques. The abdominal route can be done laparoscopically or via laparotomy by the Huntington, Haultain, and Oejo procedures. In Spinelli and Kustner techniques, the prolapsed uterine fundus is repositioned by transecting the anterior and posterior cul-de-sac, respectively. In Huntington's procedure, the bilateral round ligaments are grasped and gently pulled upward until the uterus returns to its original position. In Haultain procedure, a longitudinal incision is made over the constriction ring posteriorly and traction is applied until the uterus is repositioned. Oejo's procedure also includes longitudinal incision and traction, but it is done anteriorly on the constriction ring. The uterus is then repaired in two or three layers. The abdominal route is preferred over the vaginal route because of better visualization, more precise incision of the constriction ring, easier repositioning and traction on the round and broad ligaments, more adequate hemostasis, and allows for proper suturing.<sup>[1]</sup> As for this case, Huntington and Haultain procedures were attempted laparoscopically. But due to the considerable firmness of the constriction ring, both were aborted, thus the need to proceed to laparotomy.

Based on several reported cases, patients would often seek delayed consultation ranging from 6 months to 1 year, which may inevitably cause tightening and increased rigidity and firmness of the constriction ring. When this happens, Johnson's maneuver will likely be unsuccessful and laparoscopic approach will not be the most ideal.

In young primiparas, fertility preservation is a must. The patient and family were counseled regarding the need to delay succeeding pregnancy and the necessity to undergo cesarean section for future deliveries cannot be overemphasized.

## Conclusion

The techniques and maneuvers mentioned above have all been established and tested in this age and time. The management of uterine inversion varies, and the most effective available surgical technique in repositioning chronic uterine inversion is identified on a case-to-case basis. The outcome of the different repositioning methods is greatly affected by the timing of detection of uterine inversion. It is for this reason, the World Health Organization and Department of Health emphasized

the importance of regular prenatal checkups, antenatal surveillance, and the need for proper management of the third stage of labor by delivering in hospitals assisted by medical professionals to decrease maternal morbidity and mortality. Despite these mandates, there are still patients coming in due to uterine inversion and they usually present with severe anemia. This paper therefore recommends empowering awareness on the importance of prenatal and antenatal care, and complications of mismanaged third stage of labor for earlier detections, and prevention of morbidities such as severe anemia and the need for surgical interventions.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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