

Case Report

Stepwise Surgical Management of Chronic Puerperal Uterine Inversion

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Abstract: Background: Acute uterine inversion is a life-threatening obstetrical emergency. If not corrected properly at the time of recognition, acute uterine inversion may then progress to a chronic condition. Case: A 31-year-old P1 s/p spontaneous vaginal delivery 8-weeks ago presented with persistent vaginal bleeding and pelvic pressure. Her delivery was complicated by postpartum hemorrhage requiring blood transfusion, secondary to acute uterine inversion. Pelvic exam revealed a small uterus with complete uterine inversion. A stepwise surgical intervention with the least to most invasive methods was performed. Conclusion: the patient recovered well from the surgery and a normal menstruation resumed 9 weeks after surgical correction. A stepwise surgical management is ideal for patients with chronic puerperal uterine inversion, especially those of young age and desires for future fertility.

Keywords: chronic uterine inversion, puerperal uterine inversion.

INTRODUCTION

Uterine inversion is a very rare condition with incidence of 1:2,000~50,000 births. Maternal mortality secondary to puerperal uterine inversion has been reported to be as high as 15% (Dwivedi, *et al.*, 2013). Although the management is difficult because of the rare nature of the condition, there are several methods described in the literature, including pharmacologic agents, manual maneuvers and surgical interventions.

The aim of this case report is to describe a case of a stepwise surgical management of chronic puerperal uterine inversion, presented to us at 8 weeks after vaginal delivery and incomplete correction of acute uterine inversion.

CASE PRESENTATION

A 31-year-old P1001 s/p spontaneous vaginal delivery (SVD) 8 weeks ago at outside hospital (OSH) presented with persistent vaginal bleeding and pelvic pressure. She reported an uncomplicated prenatal course but her delivery was complicated by postpartum hemorrhage (PPH) requiring blood transfusion, secondary to acute uterine inversion.

According to OSH records, a baby was born spontaneously with birth weight of 3,089 grams and APGAR scores of 8 and 9. A uterine inversion was

recognized while delivering the placenta. The placenta was manually removed and noted to be intact. The uterus was already contracted so manual replacement of the uterus was difficult. They moved the patient to the operating room, where “hydrostatic reduction with a 60 milliliters (ml) Foley balloon and vaginal packing” was performed to manage her uterine inversion. Total estimated blood loss was noted to be 3,500 ml. She received a total of 6 units of packed red blood cells, 2 units of fresh frozen plasma and Tranexamic acid during the hospital stay. Her pre-delivery hematocrit (HCT) was 36.7%, and then it ranged 17.9-21.1% after PPH episode. On postpartum day (PPD) #4, she was discharged home after Foley balloon and vaginal packing were removed. Patient reported that her vaginal bleeding persisted thereafter. On PPD#47, her provider placed her on medroxyprogesterone 10 milligrams (mg) daily by mouth, which helped to slow down vaginal bleeding. On PPD#50, MRI of pelvis was performed (see Figure 1a), which showed complete uterine inversion.

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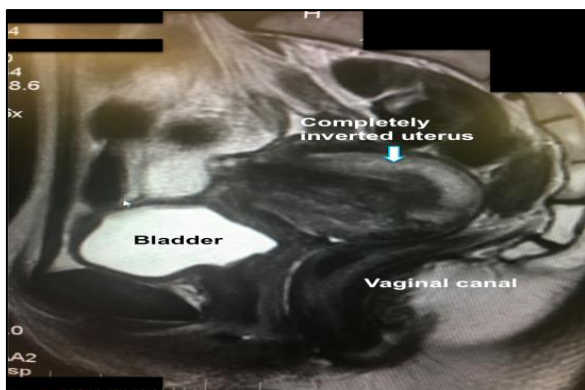


Figure 1a. MRI of pelvis on PPD# 50 showing complete uterine inversion in the vaginal canal. Uterine endometrial lining noted to be 1.5 centimeters. Cervix was difficult to delineate and both ovaries appeared to be normal. No free fluid in the pelvis.

Her provider told her she needs hysterectomy: she came to our clinic for second opinion because the patient strongly desires for future pregnancy. Her past medical and surgical histories were non-contributory. Physical exam noted a thin White female without acute distress with normal vital signs: Blood Pressure: 120/81 mmHg, Pulse: 77 bpm, Weight: 47.2 Kg, Height: 155 centimeters (cm). Pelvic exam revealed a small uterus with complete uterine inversion (see Figure 1b).

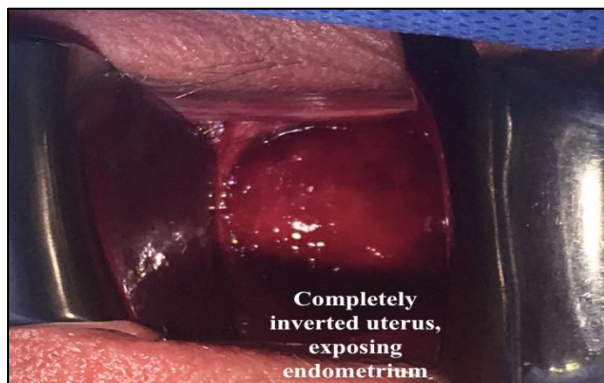


Figure 1b. Pelvic exam revealed a small uterus with complete uterine inversion: a 4cm x4cm circular area of endometrium exposed to the vagina, cervix unable to delineate, no active bleeding from the endometrium lining.

Treatment and Management

A stepwise surgical intervention was planned starting with the least to most invasive methods under general anesthesia: 1) vaginal reduction of uterine inversion with uterine relaxation with Nitroglycerine IV titration, 2) laparoscopic reduction of uterine inversion with Hungtington method and uterine relaxation with Nitroglycerine IV titration, 3) creation of a small hysterotomy to aid the reduction laparoscopically, 4) reduction of uterine inversion via mini-laparotomy and 5) cystoscopy. The patient was consented with a witness present after extensive discussion of risks, benefits and alternatives.

Procedure Details: Cefazolin 2 grams and Metronidazole 500 mg were given intravenously (IV) for antibiotic prophylaxis. After giving 50 micrograms (mcg) of IV Nitroglycerine for uterine relaxation, vaginal reduction was attempted but failed. Decision was made to proceed to the laparoscopy. Utilizing a common laparoscopic technique, the abdomen was entered without difficulty after the pneumoperitoneum was created with CO2 gas. The patient was placed in Trendelenburg position. Laparoscopic surveillance revealed that bilateral round ligaments and proximal portion of bilateral adnexa were completely involuted through the cervix showing classical flower vase appearance (see Figure 2). Both ovaries and fallopian tubes appeared to be normal. While the inverted uterus was on traction from the vagina and titration of IV nitroglycerine, the round ligaments were gently pulled up from the central portion of inverted uterus (Hungtington's method). However, the uterus continued to stay inverted. The decision was made to release tension on the anterior portion of the constricting cervical ring. Using an electrocautery hook, a 3cm-long, full thickness midline anterior incision was made, away from the bladder, to release tension on the constriction ring, while traction was applied on the round ligaments. However, the uterus still remained inverted. At this time, decision was made to covert to open procedure: Trendelenburg position was reversed and a 4cm Pfannenstiel incision was made. After bladder flap was created, the prior anterior uterine incision was extended into the crater of the inverted uterus up to the fundus level. With traction on the round ligaments (using Babcock clamps) and upward traction (by vaginal hand inside uterine body), the uterus was replaced back into the normal position. No trauma on endometrial cavity/round ligaments/adnexa was noted. The endometrial, myometrial, and serosal layers were reapproximated separately in a running fashion using a reabsorbable suture. Excellent hemostasis and uterine tone were noted (uterus size noted to be 6 weeks). Cystoscopy revealed grossly normal bladder mucosa (without cystotomy/suture/stone/lesions), bilateral ureteral jets and normal urethra. After adhesion barrier sheet was placed over the uterine incision, the abdominal layers and trocar sites were closed in a usual fashion.

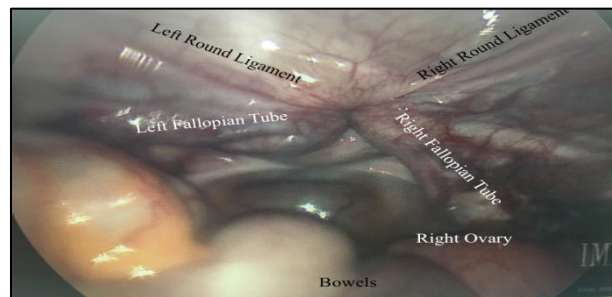


Figure 2. Intraoperative finding of complete uterine inversion: bilateral round ligaments and proximal portion of bilateral adnexa were completely involuted through the cervix showing classical flower vase appearance.

Outcome and Follow Up

On postop day (POD)#1, patient tolerated food well, voiding freely, ambulating and pain was well controlled with by mouth medications. Bedside transabdominal sonogram confirmed the proper position of uterus. Patient was discharged home on POD#1 with doxycycline 100mg by mouth for 10 days. On POD#13, the uterus and cervix appeared to be normal (see Figure 3). Patient resumed her normal menstruation 9 weeks after surgical correction. Patient was strongly informed that she would need careful follow-ups during future pregnancies for potential risk of uterine ruptures occurring during a future pregnancy.



Figure 3. Postoperative follow up: uterus and cervix are in the correct position

DISCUSSION

Acute uterine inversion is a life-threatening obstetrical emergency, which may lead to maternal death if not recognized and treated promptly. If not corrected properly at the time of recognition, acute uterine inversion may then progress to a chronic condition, as seen in our patient who presented to us 8 weeks after delivery.

Although the exact cause of uterine inversion is unknown, the common predisposing factors of puerperal uterine inversion include spontaneous (short umbilical cord, sudden rise of intra-abdominal pressure, placenta abnormality, connective tissue disorders) and iatrogenic (due to mismanagement of 3rd stage of labor). The patient with chronic uterine inversion can present with vaginal bleeding, vaginal mass, lower abdominal/pelvic/lower back pain, foul-smelling vaginal discharge and/or possible urinary disturbance (Rudra, *et al.*, 2011).

The management of acute uterine intervention includes treatment of hypovolemic shock and manual manipulation of the uterus (Johnson maneuver) plus-minus use of pharmacologic agents (i.e. nitroglycerin, magnesium sulfate or terbutaline) to assist in uterine relaxation for achieving correction.

However it is known that surgical intervention is necessary in chronic uterine inversions because the walls of the chronically inverted uterus are completely

involved with retraction and little elasticity. Therefore the constricting ring and inelastic walls have to be overcome along with the rigidity of the retaining myometrium, which cannot be overcome (Haultain, 1908).

The traditional surgical options for chronic uterine inversion include abdominal approaches (Huntington's and Haultain's) and vaginal approaches (Spinelli's and Kustner's). Huntington method involves grasping the round ligaments and the uterus below the area of inversion and slowly pulling up repeatedly until the uterus is reinverted. Haultain method involves incising posterior of the vaginal-cervical ring and carrying up the posterior wall of the uterus until it is reinverted to its normal anatomy. Compared to Spinelli method involving an incision on the anterior aspect of the cervix and then the uterus is reinverted, Kustner method is to enter the pouch of Douglas vaginally and to split the posterior aspect of the uterus and the cervix for reinverting the uterus (Birge, *et al.*, 2015). Some case reports also added a possibility of utilizing laparoscopic assisted vaginal approach to correct acute puerperal and chronic uterine inversion (Vijayaraghavan&Sujatha, 2006 and Zhang, *et al.*, 2015).

In our case, we approached the case with a stepwise surgical management utilizing the least invasive to most invasive methods (vaginal → laparoscopic-assisted vaginal → abdominal) to achieve a successful correction of uterine inversion and preserving the uterus for future fertility.

We utilized IV nitroglycerine titration as a uterine relaxing agent. While terbutaline takes about 2 minutes to take effect and magnesium sulfate takes about 10 minutes to be effective (Beringer&Patteril, 2004), the onset of action for IV nitroglycerine takes only 75-95 seconds and the side effects are minimal with low doses (Saroa, *et al.*, 2013). Recurrence after uterine inversion correction is known to be rare in future pregnancies if proper obstetric follow-ups are planned (Baskett, 2002).

To our knowledge, this is the first case reported as uterine repositioning and preservation using a stepwise surgical management for a patient suffering from incompletely treated puerperal uterine inversion. A stepwise surgical management should be performed for patients suffering from chronic puerperal uterine inversion, especially those of young age and desires for future fertility.

Learning Points

- Acute uterine inversion is a life-threatening obstetrical emergency. If not corrected properly at the time of recognition, acute uterine inversion may then progress to a chronic condition.

- There are several methods described in the literature, including pharmacologic agents, manual maneuvers and surgical interventions.
- A stepwise surgical management should be performed for patients suffering from chronic puerperal uterine inversion, especially those of young age and desires for future fertility.

Patient Perspective

"I am extremely thankful and grateful to Dr. Chong and her team of associates that were finally able to give me the feeling that I was in expert hands. From our first appointment, and subsequent correspondence I was confident that my case was reviewed in detail and I had a team of medical professionals that truly had the knowledge and expertise to handle my case.

After a total of 27 hours of labor, five of those hours spent pushing, I delivered a healthy baby girl and minutes after was rushed to the operating room as my uterus inverted during the delivery of the placenta. The whole ordeal was frightening, to say the least. When I was discharged from hospital, I had no sonogram. During my postpartum and postoperative period, I experienced constant bleeding which never subsided with time. I made many phone calls and had appointments to my OBGYN to address my bleeding. I was told if I wasn't soaking a pad every hour, it was normal. I called three more times in the subsequent weeks because of my bleeding. I was finally prescribed progesterone to slow the bleeding. At my 8 week follow up when I was scheduled to return to my first grade teacher position in two days, they discovered my uterus was still inverted through an internal sonogram and a lot of bleeding. I was told I would have another surgery but if she was not able to correct the uterus, they would have to perform an immediate hysterectomy due to bleeding.

I then sought for a second opinion. My records were sent and from my first appointment with Dr Chong, I was confident, she had reviewed my case and had a detailed plan to present. She spent a great deal of time explaining the procedure and then surgery was scheduled. It was a five-hour drive for me and the hospital was accommodating to the distance I had to travel. After the operation, while I was in recovery, Dr. Chong explained in detail to my husband and mothers

the procedure they used to correct the inversion. Dr. Chong came to my hospital room and then explained it to me. I had followed up appointments and she was very thorough with my care. She was also always available by phone. I was also impressed with a smoothly run office and friendly qualified staff. From our first meeting with Dr. Chong, I felt confident in my medical care and was so thankful I made the decision to get that second opinion."

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