

The role of sonography in the diagnosis of chronic puerperal uterine inversion: A case report*

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ABSTRACT

Chronic puerperal uterine inversion is a rare and life-threatening obstetric emergency which requires emergent treatment. We present a case of a 27-year-old Gravida 2 Para 2 (2002) with chronic uterine inversion. A bleeding, 4 x 4 x 5 cm fleshy knob like mass protruding from the cervix, was seen during vaginal inspection. Two-dimensional transvaginal sonography and 3-dimensional imaging clinched the diagnosis of uterine inversion. The patient underwent Haultain's procedure and was discharged improved with resumption of normal menses. Postpartum transvaginal sonography revealed a normally positioned uterus.

Keywords: chronic puerperal uterine inversion, Haultain's procedure, sonography

INTRODUCTION

Uterine inversion is a rare obstetric emergency. This postpartum complication has an academic importance due to its rarity and severity. When not immediately identified, the massive and often underestimated blood loss can lead to hypovolemic shock and maternal death that can reach 15% in some series.¹ Due to the emergent nature of these cases, prompt diagnosis and early initiation of appropriate treatment is crucial.

Majority of the cases of uterine inversion are puerperal in nature with incidence ranging from approximately 1 in 550 to 1 in 2500-7000.²⁻⁵ Non-puerperal uterine inversion, on the other hand, is extremely rare, accounting to only one sixth of all cases of uterine inversion. Most of these cases are associated with the traction effect of mass lesions, mostly benign submucous myomas, attached to the fundus.

According to the Annual Report of the Philippine Obstetrical and Gynecological Society, there were nine reported cases of uterine inversion in 2015, 7 of which were puerperal while 2 were non-puerperal in nature.

Several case reports regarding uterine inversion have been written in literature. In our institution, three reports were written regarding non-puerperal uterine inversion, but none on puerperal uterine inversion. Lipana in 2008 reported a case of uterine inversion secondary to prolapsed submucous myoma who underwent Haultain's procedure. Anzures in 2008 reported two cases of non-

puerperal uterine inversion both due to prolapsed submucous myoma. In 2013, Seneris reported a case of an 18-year-old nulligravid with complete uterine inversion with no apparent cause.

Puerperal uterine inversion is an obstetric emergency which requires rapid diagnosis and immediate clinical intervention. Recognition and appropriate interpretation of typical sonographic findings would help clinch the diagnosis of uterine inversion potentially improving clinical outcome.

The aim of this article is to describe the sonologic characteristics of uterine inversion which would clinch our diagnosis hence avoiding delays in the management. There are several therapeutic strategies described in the literature, including drugs, manual maneuvers and surgical interventions.

CASE

This is a case of A.A., a 27-year-old Gravida 2 Para 2 (2002), single, from Laguna, who consulted for a three-year history of vaginal bleeding.

She has no hypertension, diabetes, asthma, no history of thyroid disease nor blood dyscrasias and she has no allergies to food and medications. She has no known heredo-familial diseases.

She is a high school graduate and currently works as a factory worker. She has no vices. Her first coitus was at 20 years-old with one lifetime non-promiscuous sexual partner. She has no history of hormonal contraceptive and intrauterine device use or affliction with sexually transmitted infections.

She had her menarche at 16 years-old with subsequent menses occurring at regular monthly intervals, lasting for 5 days using 2 pads per day with no episodes of dysmenorrhea.

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She is a Gravida 2 Para 2 (2002). Both pregnancies were carried to term, born via spontaneous vaginal delivery assisted by a traditional birth attendant. Both children are alive and well. Her first pregnancy had an unremarkable course. Her second pregnancy was in 2012, 3 years prior to this admission. Her second pregnancy was delivered at home by a traditional birth attendant. She had an unremarkable course of labor. However, during the delivery of the placenta, a bulging mass was seen prolapsing out of the introitus. The placenta, which was attached to the mass, was easily removed. The prolapsing mass, which seemed to be the inverted uterus, was replaced back into the vagina after detachment of the placenta. The patient was stable postpartum with minimal amount of vaginal bleeding. Two weeks postpartum, she reported vaginal bleeding, using up 2-3 diapers per day, lasting for two weeks. She did not seek consult during this time.

During the interim, subsequent menses increased in amount and duration, using 5 pads per day and lasting up to 7 days. She also noted intermenstrual spotting and postcoital bleeding. Persistence of the vaginal bleeding prompted consult at a local tertiary hospital where she was diagnosed with cervical myoma and advised hysterectomy. The patient, however, was lost to follow up. One month after her initial consult, the patient consulted another hospital where she was subsequently admitted. The admitting impression was cervical myoma and anemia secondary to chronic blood loss. She was transfused with 3 units of packed RBCs and was advised hysterectomy. The patient, however, went home against medical advice.

Two weeks prior to this admission, the patient sought consult at the Out-patient Department of a tertiary hospital for second opinion. During this time she had vaginal bleeding, easy fatigability and dizziness.

On review of systems, there was no fever, headache, cough, colds, dyspnea, urinary or bowel movement changes. She was conscious, coherent, ambulatory, and not in cardiorespiratory distress. She had stable vital signs with BP of 100/70, HR 80, RR 20, Temperature of 36.5°C. She had pink palpebral conjunctivae, anicteric sclerae, no anterior neck mass, no cervical lymphadenopathy. She had equal chest expansion, clear breath sounds with no crackles nor wheezes noted. She had a dynamic precordium with distinct heart sounds, normal rate and regular rhythm. There were no murmurs appreciated. She had full and equal pulses, with pink nail beds and had no edema, cyanosis or clubbing. Her abdomen was flat, soft and non tender.

On speculum examination, there was a 4 x 4 x 5 cm fleshy, knob-like mass occupying the upper one-third of the vaginal canal (Figure 5). The cervix was not seen. The vaginal walls were smooth. On internal examination, the

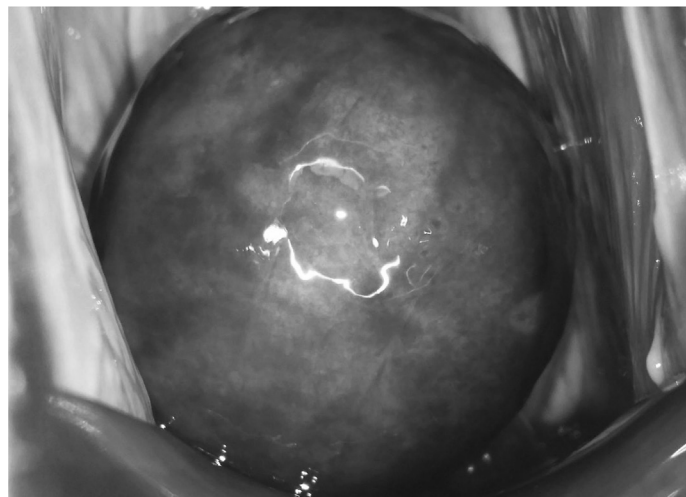


Figure 5. Speculum examination revealing a 4 x 4 x 5 cm fleshy, knob-like mass occupying the upper one-third of the vaginal canal. The cervix was not seen.

cervix was dilated and effaced by a 4 x 4 x 5 cm fleshy knob-like mass which seems to be attached by a broad-based pedicle to the anterior cervical stroma. The corpus was assessed to be small, with no adnexal masses and tenderness.

The initial assessment was cervical myoma. Transvaginal ultrasound was performed, which revealed that the cervix was inverted and effaced measuring 1.6 X 0.8 cm anteriorly and 2.1 x 0.5 cm posteriorly, with homogeneous stroma. The inverted uterine corpus measured 5.3 x 3.8 x 3.7 cm with note of a pseudostripe representing the interface of the two opposing serosal surfaces. The myometrial echogenicity was homogeneous from the isthmic up to the fundal area. Color flow mapping of the inverted corpus showed inverted uterine vessels spanning through the whole uterine length. The right ovary measured 2.2 x 2.5 x 1.4 cm. The left ovary measured 1.6 x 2.0 x 1.1 cm. There were no adnexal masses seen. There was no free fluid in the cul de sac. The sonographic findings were consistent with uterine inversion (Figures 1-4).

The patient was subsequently admitted for further management. Complete blood count showed anemia with hemoglobin of 66 and hematocrit of 0.27. Other laboratories were normal. The patient was transfused with two units of packed RBC and subsequently underwent Haultain's procedure to reposition the uterus back into the abdominopelvic cavity.

Intraoperatively the uterine corpus was inverted from the level of the midcorpus with a constriction ring noted at the lower segment. Both ovaries and fallopian tubes were pulled along with the uterus (Figures 6-7).

The patient tolerated the procedure well. She remained afebrile and asymptomatic throughout her hospital stay. Change of dressing was done on the second

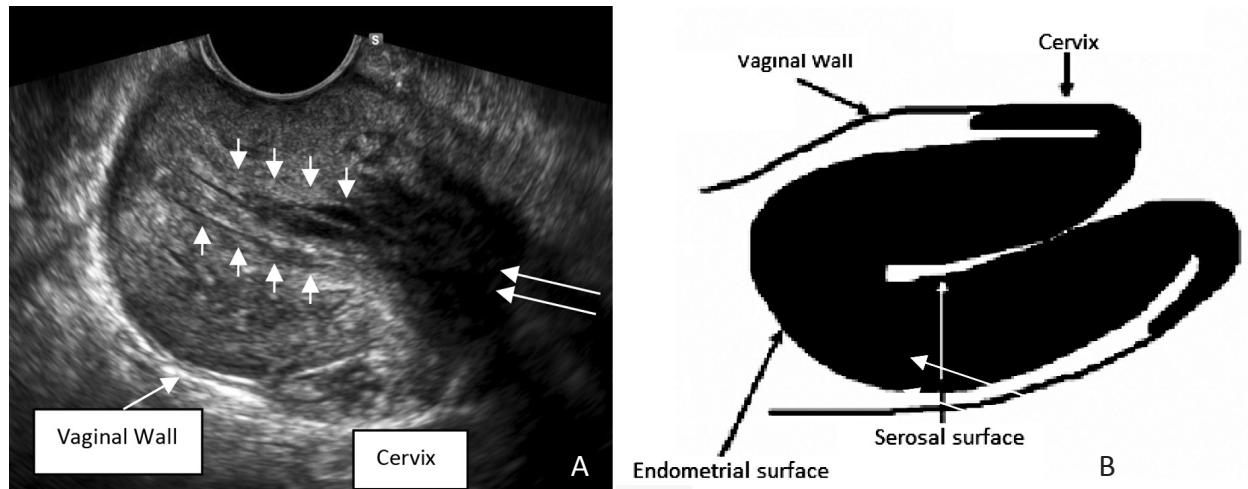


Figure 1. A. Mirror reflection of the normally positioned uterus projecting into the vagina. Central cleavage (long arrows) represents path of inversion. A pseudo-stripe was present (short arrows) representing the interface of the two opposing serosal surfaces mimicking an endometrial stripe. B. Illustration of the pseudo-stripe (Illustration from Rana et. al., 2009)



Figure 2. On color Doppler, the pseudo-stripe revealed inverted uterine vessels spanning the entire uterine length.

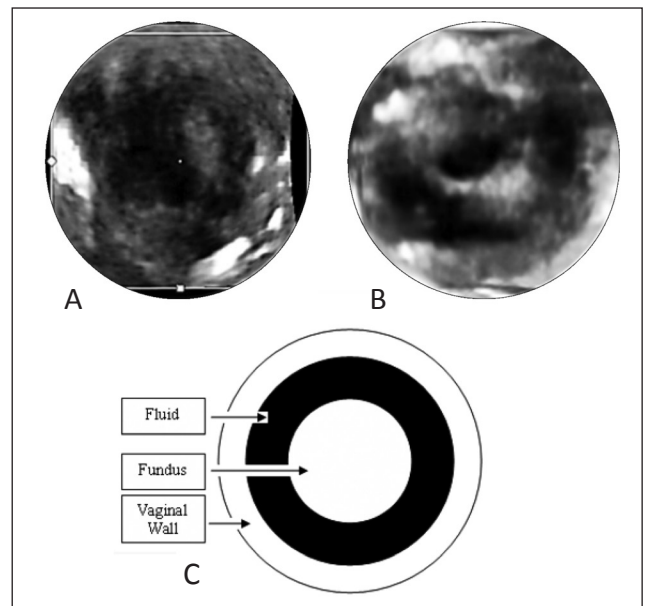


Figure 3. A. Transabdominal transverse sonogram showing the target sign with the hyperechoic inverted fundus centrally surrounded by hypoechoic fluid between the fundus and vaginal wall. The image plane is in the lower pelvis. B. 3 dimensional sonogram of the target sign. C. Illustration of the target sign. (Illustration from Rana et. al., 2009)

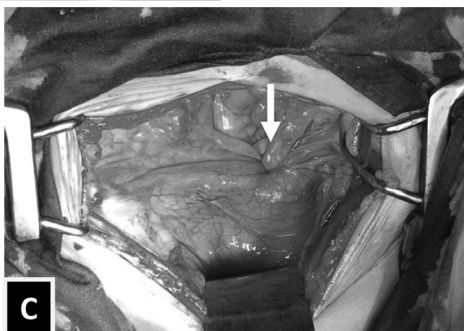
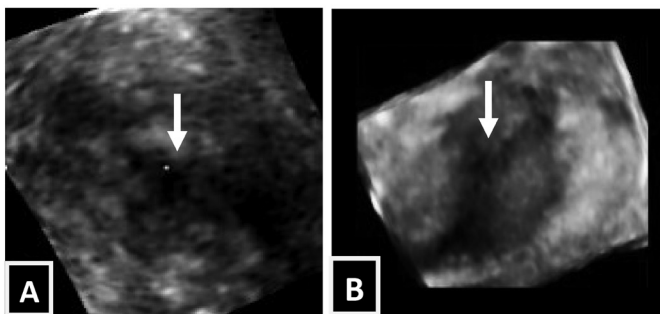


Figure 4. A. A transverse transabdominal sonogram at the level of lower segment shows X-shaped hypoechoic center corresponding to folded endometrial cavity. B. 3 dimensional sonogram of the X-shaped hypoechoic center. C. Intraoperative findings of the X-shaped hypoechoic center, indicating the invaginated uterus. (Arrow representing the path of inversion)



Figure 6. Repositioned uterus after Haultain's procedure. Sutures are seen at the posterior myometrial wall.

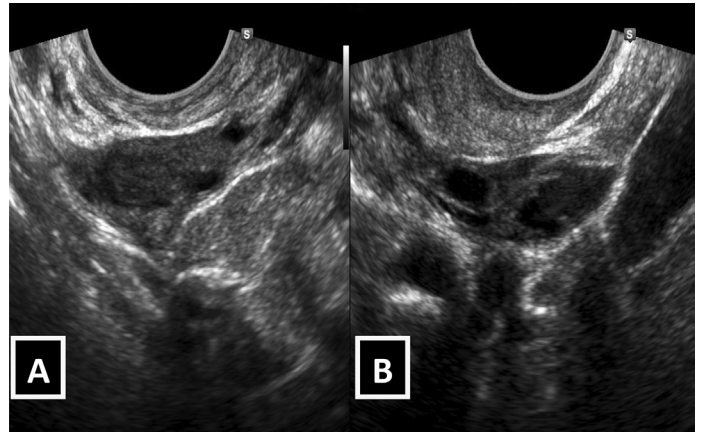


Figure 7. Transvaginal images showing the bilateral ovaries with suspensory ligaments pulled along the with the uterus within the vaginal canal. (A: Right ovary, B: left ovary)



Figure 8. Transvaginal sonography of the normally positioned anteverted uterus with multiple echogenic foci (arrows) at the posterior myometrial wall suggestive of surgical sutures, one week after surgery



Figure 9. Transvaginal sonography of the normally positioned anteverted uterus with multiple echogenic foci (arrows) at the posterior myometrial wall suggestive of surgical sutures, one month after surgery

postoperative day. The wound was dry and intact with no erythema, discharge or hematoma. She was discharged stable on the fifth postoperative day.

One week postoperatively, transvaginal sonography revealed a normally positioned, anteverted uterus with smooth contour and homogeneous echopattern measuring 8.4 x 4.6 x 3.2 cm. Adhesions were noted at the posterior uterine wall. The cervix measured 3.3 x 3.1 x 2.3 cm, with homogeneous stroma and distinct endocervical canal. The endometrium was hyperechoic measuring 0.2 cm with intact subendometrial halo. There were multiple echogenic foci seen from the posterior isthmic to the midcorpus, suggestive of suture tracts. The right ovary measured 2.6 x 1.9 x 1.2 cm, while the left ovary measured 3.1 x 1.9 x 1.5 cm. There were no adnexal masses seen (Figure 8).

Repeat transvaginal sonography one month after the surgery revealed normal findings (Figure 9).

DISCUSSION

Uterine inversion is defined as the passage of the uterine fundus through the endometrial cavity and cervix, thereby turning the uterus inside out.² It can be classified into puerperal, which constitutes 95% of cases and non-puerperal, in 5% of cases.

The reported incidence of uterine inversion varies considerably in the literature, with reports ranging from approximately 1 in 550 to 1 in 2500-7000.³⁻⁵ Puerperal uterine inversion, which was seen in our index patient, is a rare complication of vaginal delivery which if not recognized and treated immediately, can lead to life-threatening bleeding and shock. Maternal mortality has been reported to be as high as 15%.¹

Non-puerperal uterine inversion, on the other hand, is extremely rare, developing mostly after 45 years of age, and accounting to only one sixth of all cases of uterine

inversion.^{4, Anzures, 2008} This condition can be seen due to gravitational mass effect of tumors. Causes include mostly benign conditions such as leiomyomas, and rarely uterine tumors such as sarcomas and carcinomas.⁵

According to the Annual Report of the Philippine Obstetrical and Gynecological Society, there were nine reported cases of uterine inversion in 2015, 7 of which were puerperal while 2 were non-puerperal in nature.

A case report written by Anzures in 2008, stated that the pathophysiology of inversion entails that a portion of an otherwise normal uterine fundus indent towards or prolapses into the cervical canal. She further stated that a degree of uterine relaxation combined with a simultaneous downward movement of the fundus, followed by the resumption of uterine contractions at precisely the right moment, must occur to result in an inversion.⁶ Most authors would attribute the cause of puerperal uterine inversion to excessive traction on the umbilical cord during delivery. A fundally located placenta, rapid uterine emptying and nulliparity are other predisposing factors for uterine inversion.^{2,7} However, the exact cause of complete uterine inversion is still unexplained. In fact, there are several cases in which no tension was carried out on the cord. Other risk factors associated with this situation are fetal macrosomia, excessive fundal pressure, placenta accreta, short umbilical cord, use of Credes method during manual removal of placenta, ligaments laxity, and congenital abnormalities of the uterus.^{2,3}

The process of uterine inversion usually starts in the fundus with variable degree of inversion. Rana, et. al in 2009, classified complete puerperal uterine inversion anatomically or chronologically. The anatomic classification is described in 4 stages: Stage 1, the inverted fundus extends to but not through the cervix; Stage 2, the inverted fundus extends through the cervix but remains within the vagina; Stage 3, the inverted fundus extends outside the vagina; and Stage 4, total uterine inversion along with vaginal inversion occurs.⁷ Chronologically, uterine inversion can be classified based on the delay between the delivery and diagnosis of uterine inversion, defined as follows: Acute, occurring within 24 hours of delivery; Subacute, occurring 24 hours to 30 days postpartum; and Chronic, occurring greater than 30 days postpartum.⁶ On the basis of the clinical history and sonographic findings, our patient had chronic stage 2 uterine inversion.

The diagnosis of uterine inversion is clinical. It is an obstetric emergency and a diagnostic challenge in gynecology. Its presentation can vary with various degrees and rates of inversion. There may be only pain when it is acute and hemorrhage or unexplained shock when it is severe. However, only minor leucorrhoea or irregular hemorrhage may be seen in chronic inversion, as seen in our case. Chronic uterine inversion must be kept in mind in

cases of periodical vaginal bleeding and/or fragile bleeding tissues in the vagina.⁸ Puerperal uterine inversion is often suspected in the presence of massive blood loss after childbirth or in the absence of uterine fundus during abdominal palpation. Hypotension and tachycardia may supervene and evolve into hypovolemic shock. The morbidity and mortality associated with uterine inversion correlate with the degree of hemorrhage, rapidity of diagnosis, and the effectiveness of the treatment. In acute uterine inversion, massive blood loss with hemorrhage could be seen and hypovolemia should be vigorously treated with fluid and blood replacement. Although less common, inversion may present as a mass on physical examination, such as in our patient. In this setting, the correct diagnosis can be easily delayed, which in turn can lead to subacute or chronic inversion and increase the need for surgical intervention.⁷ Our index patient was diagnosed with cervical myoma for which a hysterectomy was advised. This incorrect diagnosis has led to the delay in the management of our case.

The observation of the uterine fundus beyond the vaginal introitus in the complete form or the palpation of the fundus through the external os in the 3rd degree uterine inversion is the most common sign. When a physical examination is inconclusive, such as in our case, and the patient is hemodynamically stable, sonographic evaluation is critical in clinching the diagnosis of uterine inversion. Sonographic findings are striking and can be easily understood if the pathologic process of the uterine inversion is known.

Several studies have been published, describing the sonographic characteristics of uterine inversion. Rana, et. al cited three (3) distinct sonographic characteristics of the inverted uterus, which would help us arrive at the correct diagnosis. First, is a sonographic finding of a mirror reflection of the normally positioned uterus projecting into the vagina. Second, a pseudostripe can be seen because the interface of the two opposing serosal surfaces mimics an endometrial stripe. Partial uterine inversion was also described with the appearance of a Y-shaped endometrium. Third, 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.⁶ These three Sonographic characteristics were seen in our patient using 2-dimensional gray scale and 3-dimensional transvaginal and transabdominal sonography (Figures 1-3). Furthermore color Doppler of the pseudostripe revealed inverted uterine vessels spanning the entire uterine length (Figure 2).

Momin et, al in 2009, described the sonographic findings of uterine inversion from acute to chronic form. The initial sonographic examination of acute uterine

inversion shows bulky uterus with fundal inversion. A central cleavage represents path of inversion. A transverse transabdominal sonogram at the level of lower segment shows X-shaped hypoechoic center corresponding to folded endometrial cavity and the path of inversion.³ This was also seen in our patient during a 3-dimensional scan done preoperatively (Figure 4). Transvaginal longitudinal sonogram can also show partial inversion of uterus which reaches the cervical region (Figure 1).

Longitudinal transabdominal sonogram of a chronically inverted uterus would later show a miniature copy of a previously seen inverted uterine fundus. Transverse transabdominal sonogram would show both ovaries drawn close to the uterus due to the inward pull. Transverse transabdominal sonogram of the uterus in chronic stage would also show a rounded echogenic inverted fundus in the lower segment surrounded by uterine walls.³

Identifying the location of the ovaries in relation to the inverted uterus is another key sonographic finding in the recognition of uterine inversion. The ovaries may appear attached to each other and retracted to the midline especially in cases of complete uterine inversion.⁹ The ovaries may also be seen at the sagittal plane of the uterine indentation giving the appearance of a "stuffed olive".¹⁰ This sonographic characteristic was not seen in our patient. However, we noted that the suspensory ligament were pulled along with the uterus within the vaginal canal.

Once uterine inversion is diagnosed, attempts to reduce it should be made by methods described in obstetric practice. If diagnosed early, immediate manual vaginal replacement along with anesthesiologist assistance and medical management is the best option. Cases not treated on time may require surgical intervention, especially in chronic cases to open up the dense constriction ring.

The management of uterine inversion should be step-wise, involving both non-surgical and surgical approaches.¹¹ Calling for appropriate help is foremost in the management, followed by resuscitation and stabilization of the patient. Once the diagnosis has been established, an immediate attempt should be made to

replace the uterus manually through the vagina past the cervical ring, with pressure directed upwards towards the umbilicus. This is commonly referred to as Johnson's method. Should this fail, the next step in the management is hydrostatic replacement with the use of a 6 cm silastic ventouse cup to correct the uterine malposition. Antonelli et. al. in 2006, have described a new technique of using a silastic cup at the time of laparotomy. Use of an intrauterine Rusch balloon catheter (Modified hydrostatic method) has also been reported.¹¹

When manual reduction fails, surgical procedures are indicated. In a chronically inverted uterus, such as in our case, there is already formation of the constriction ring by the myometrium encircling the prolapsed part of the uterus, hence surgical management is employed. There are several approaches in the surgical management of uterine inversion. Huntington procedure involves a laparotomy to locate the cup of the uterus. Allis forceps is used to gently apply upward traction until the inversion is corrected. Haultain technique involves a longitudinal incision on the posterior cervical ring and reversal by gentle traction as in the Huntington procedure. A modified laparoscopic reduction has also been reported.¹¹

For our index case, a Huntington procedure was initially attempted. However, there was difficulty in performing the procedure due to a constriction ring. A Haultain's procedure, was then successfully employed to reposition the uterus.

Recurrence is rare in cases where the uterus is preserved, such as in our index case. Haultain also reported good pregnancy outcomes following the correction.¹²

CONCLUSION

Puerperal uterine inversion is an obstetric emergency which requires rapid diagnosis and immediate clinical action. Its rarity leads to limited experience in its management. The morbidity and mortality associated with this complication can be decreased by prompt diagnosis and early initiation of suitable treatment. Recognition and appropriate interpretation of typical sonographic findings would help clinch the diagnosis of uterine inversion potentially improving clinical outcome. ■

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