Evolution of a cesarean scar pregnancy into a placenta accreta at term: A case report*

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ABSTRACT

This is a case report of a first trimester cesarean scar pregnancy (CSP) evolving into a placenta accreta at term based on the ultrasound imaging. The gestational sac, initially implanted at the site of previous scar, grew into the uterine cavity as the pregnancy progressed and resulted into a viable birth complicated by placenta accreta.

Cesarean scar pregnancy is a rare form of ectopic pregnancy and is associated with increased maternal morbidity and mortality. Thus, early recognition of the salient sonographic findings is crucial because a delay could lead to a life threatening condition. Early diagnosis also gives women the option to choose between expectant management and termination of pregnancy. The exact incidence of CSP has not been determined but its incidence is on the rise in parallel with the high rate of cesarean sections. There are two types of CSP. The first type is due to the implantation of the gestational sac on the scar with progression towards the uterine cavity. In this type expectant management is justifiable since pregnancy may progress into a viable pregnancy. The second type involves growth of gestational tissues towards the bladder and abdominal cavity and is associated with uterine rupture if immediate intervention is not undertaken. In this report, we present a case of a first trimester CSP that was managed expectantly and developed into placenta accreta at term.

Keywords: Cesarean scar pregnancy, ectopic pregnancy, placenta previa, accrete

INTRODUCTION

A cesarean scar pregnancy occurs when a pregnancy implants on a cesarean scar or in a "niche" in the faulty anterior uterine wall. The main cause is still unclear but the most acceptable theory is that the blastocyst enters into the myometrium through a microscopic dehiscent tract. This may be created during the previous operations such as cesarean section, gynecologic surgery or even following manual removal of placenta. Other risk factors identified are a history of uterine infections such as endomyometritis, and a brief interval between uterine surgery and subsequent conception.[1]

The early diagnosis by transvaginal ultrasound is the gold standard in the management of CSP. It directs therapy and improves outcomes by allowing preservation of the uterus and future fertility. Also, it allows elective informed choice of treatment. Early diagnosis is possible by early clinical suspicion and an early recognition of sonographic findings.

Transvaginal sonographic criteria for diagnosis of CSP include the following: (1) visualization of an empty uterine cavity and empty endocervical canal, (2) detection of the placenta and/or a gestational sac embedded in hysterotomy scar, (3) in early gestations (</ 8 weeks), a triangular gestational sac that fills the niche of the scar or at >/ 8 weeks post menstrual week this shape is become rounded or oval, (4) a thin (1-3 mm) or absent myometrial layer between the gestational sac and the bladder, (5) a closed and empty cervical canal, (6) the presence of embryonic /fetal pole and/or yolk sac with or without heart activity, (7) the presence of prominent and at times rich vascular pattern at or in the area of a cesarean scar in the presence of a positive pregnancy test.[2] All of these criteria had to be present to diagnose CSP. Magnetic resonance imaging (MRI) is a valuable troubleshooting tool when sonography is equivocal or inconclusive

CASE

The index case is P.G., 27-year-old woman, gravida 4 para 3 (0302), married, Roman Catholic from Bulacan, who presented at the OB-admitting section after a positive pregnancy test and vaginal spotting. She has no known comorbidities. Her family as well as personal and social history were unremarkable.

She had her menarche at 12 years old occurring at regular monthly intervals lasting 3-4 days and soaking 4 pads per day with no associated dysmenorrhea. Her last normal menstruation was on March 28, 2015. She had no history of sexually transmitted disease and gynecologic surgery.

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She had a history of 3 prior cesarean deliveries. Her first pregnancy was an emergency cesarean delivery, 10 years ago (2005) for nonreassuring fetal status, followed by 2 repeat cesarean sections in 2009 and 2013. All were preterm deliveries and her first baby died 12 hours after birth due to prematurity (6 months).

First trimester

She presented at the OB admitting section with stable vital signs and essentially normal physical examination findings. Vaginal inspection revealed a cervix with normal appearance and a minor bleeding through a closed external os of the cervix. Internal examination was deferred at this time. Baseline laboratories were requested. (Table 1)

Table 1. Laboratory Results

Physical Analysis	**Result (Equivalent SI)**		
Color	Light Yellow		
Transparency	Clear		
Chemical Analysis			
Bilirubin:	Negative		
Urobilinogen:	Normal		
Ketone:	Negative		
Glucose:	Normal		
Albumin:	Negative		
Blood:	Negative		
PH:	6.0		
Nitrite:	Negative		
Leucocytes:	Negative		
Specific Gravity:	1.007		
Sediment Analysis			
	Results	**S.I. Unit Reference Range**	**Results**
RBC	0	0	/uL
WBC	0	0-9	/uL
Epithelial Cells	2	0-5	/uL
Bacteria	9	0	/uL
Mucus Thread	4	0-5	/uL

Her initial transvaginal ultrasound revealed a single gestational sac at the level of the uterine isthmus, with a mean sac diameter of 2.5 cm with a single embryo measuring 1.2 cm equivalent to 7 weeks and 3 days age of gestation (AOG) with good cardiac activity (FHT 144bpm) and a yolk sac measuring 0.3 cm (Figure 1). Both ovaries were unremarkable. There was significant thinning of the myometrium at the uterine isthmus but the serosa is intact. Color flow mapping showed scant vascularity. The initial impression was a single, live, intrauterine pregnancy, 7 weeks and 3 days AOG by crown rump length with consideration of placenta accreta (Figure 2). The initial human chorionic gonadotrophin (B-HCG) was >10,000.

A follow-up sonographic scan was performed at 8 weeks and 4 days AOG, by that time the vaginal spotting has stopped (Figure 3). The gestation sac was still at thelevel of uterine isthmus with significant thinning of

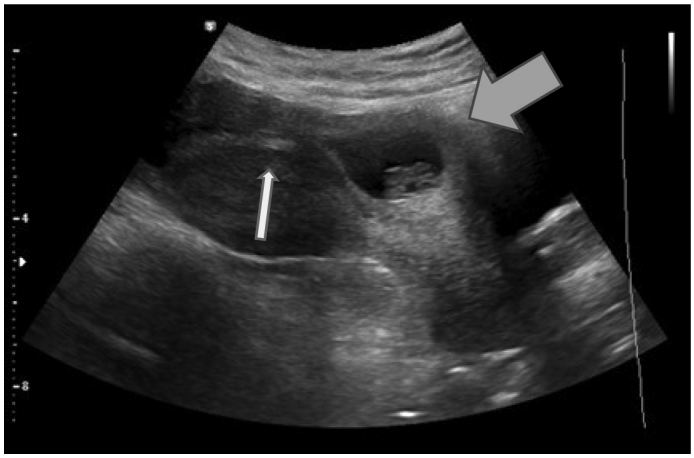

Figure 1. Transabdominal Ultrasound at 7 weeks and 3 days showing a single gestational sac at the level of the uterine isthmus. Thin arrow showed an empty endometrial cavity. The thick arrow showed anterior ballooning at the CS scar.

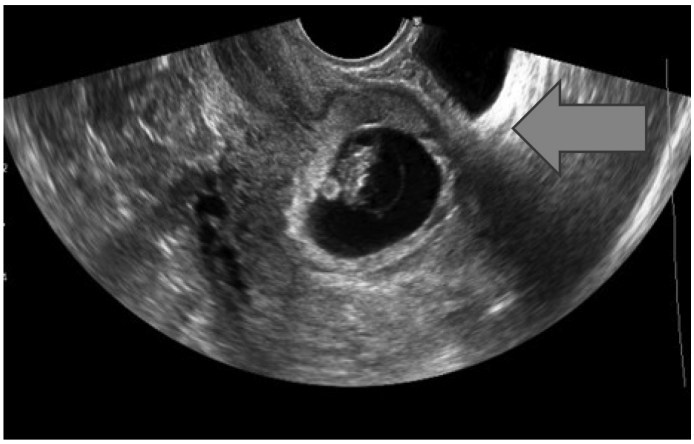

Figure 2. Transvaginal ultrasound at 7 weeks and 3 days showing an anteverted uterus with gestational sac at the anterior myometrium at the level of uterine isthmus at the site of cesarean scar.

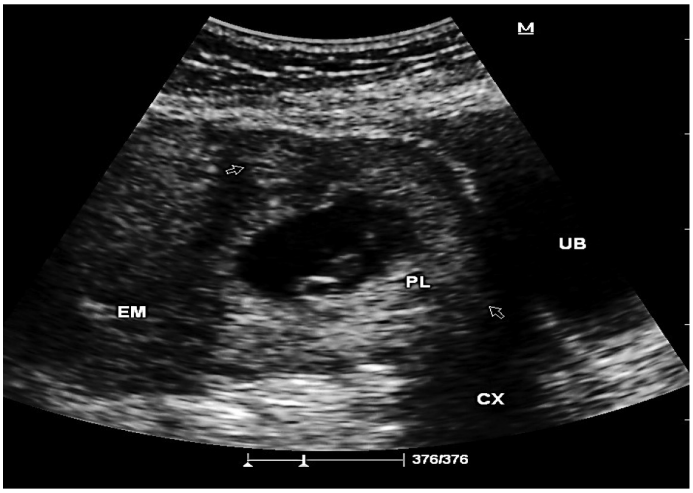

Figure 3. A follow-up transvaginal scan at 8 weeks and 4 days age of gestation, showing a gestation sac at the level of uterine isthmus with significant thinning of the myometrium. The uterine serosa was intact, with the bladder adherent to it, shows no invasion.

the myometrium. The uterine serosa was intact, with the bladder adherent to it, shows no invasion. The consideration at this time was cesarean scar pregnancy.

MRI was requested and it revealed a single gestational sac seen at the mid anterior myometrium along the region of the CS scar. The sac was not completely surrounded by the myometrium. The upper uterine body and cavity was empty and intact. The consideration was consistent with CSP (Figure 10).

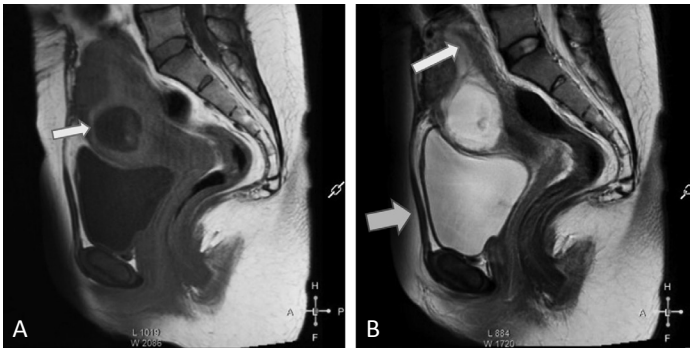

Figure 10. Magnetic Resonance Imaging Results at 8 weeks AOG. A. The GS is seen in the midanterior endomyometrium (probably along the region of the CS scar). B. The upper uterine body and cavity are intact. The adequately distended UB appear normal and has normal wall thickness (blue arrow).

Repeat ultrasound at 9 weeks and 4 days AOG revealed ballooning of anterior myometrium towards the bladder with expansion of only the lower uterine segment. The embryo was seen developing normally with good cardiac activity (Figure 4).

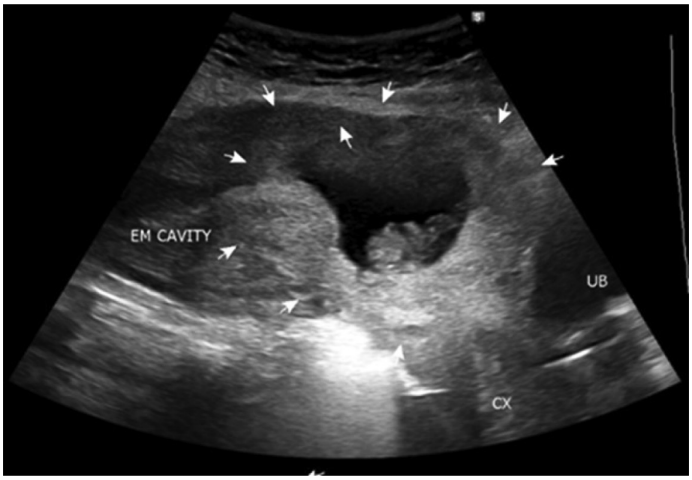

Figure 4. Transabdominal ultrasound at 9 weeks and 4 days showing ballooning of the anterior myometrium anteriorly and towards the bladder with expansion of only the lower uterine segment

In view of the above mentioned findings, the patient was informed that the pregnancy could develop into placenta previa/accreta and that she may be at risk of having major hemorrhage that would require a hysterectomy if pregnancy will be allowed to progress. After discussion, she decided to continue with the pregnancy. While admitted in the hospital, she was referred to urogynecology, thoracic and cardiovascular surgery, and perinatology services for co- management and pre-operative planning.

The pregnancy was continued without complications and the repeat abdominal scan at 13 weeks and 2 days AOG revealed that the embryo is growing normally with good cardiac activity. There was expansion of the lower uterine segment along the fetus. There was loss of retroplacental sonolucent space anteriorly but with intact and irregular uterine serosa and bladder interface. There were also placental lacunae appreciated. The consideration was CSP with placenta accreta (Figure 5). She was managed expectantly and was advised on strict regular follow -up.

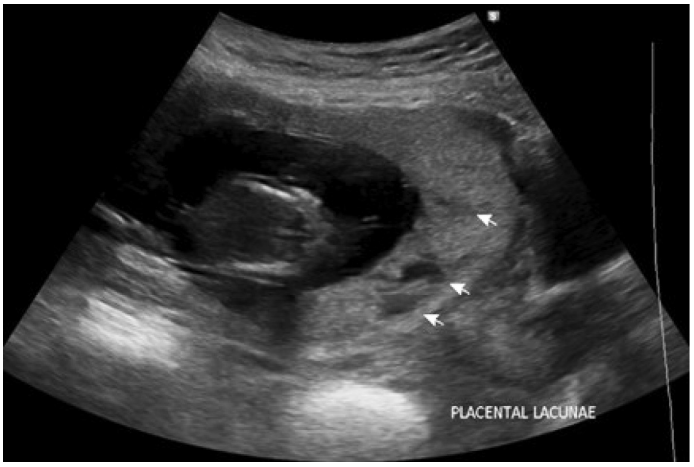

Figure 5. Transabdominal ultrasound at 13 weeks and 2 days showing expansion of the lower uterine segment. With loss of retroplacental sonolucent space anteriorly and presence of placental lacunae. The uterine serosa and bladder interface is intact.

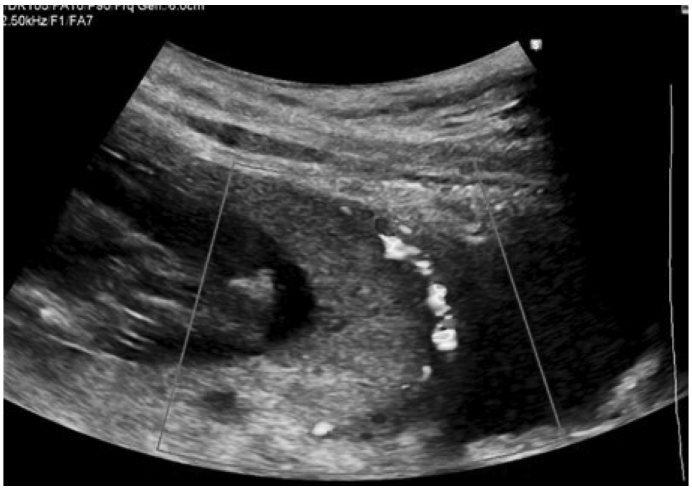

Figure 6. Transabdominal ultrasound at 13 weeks and 2 days. Color flow mapping showing hypervascularity at the uterine serosa-bladder interface.

Second trimester

Repeat MRI at 16 weeks AOG showed a markedly thinned out anteroinferior myometrial wall and no normal-looking myometrium between the uterus and urinary bladder. The placenta now is seen posteriorly compressing the endometrium (Figure 11).

On repeat scan at 17 weeks and 1 day, there was still ballooning anteriorly and expansion of only the lower uterine segment (Figure 7). The uterine serosa was still intact. The placenta was seen totally covering the os. The findings were consistent with cesarean scar pregnancy with placenta previa and accreta.

At 25 weeks and 2 days, both the 2D-gray scale scanning and color Doppler evaluation were consistent with placenta previa and accreta with the possible percreta (Figure 8). There was thinning of the uterine serosa-bladder wall complex and elevation of the tissues beyond the uterine serosa suggestive of possible bladder invasion.

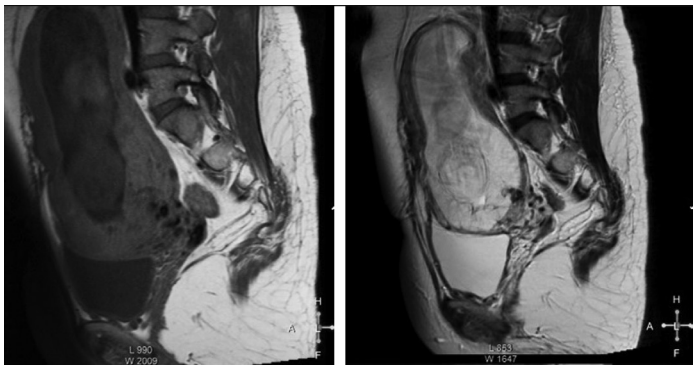

Figure 11. Magnetic Resonance Imaging Results at 16 weeks AOG. The myometrium of the anteroinferior wall of the uterus appears to be markedly thinned, with no normal-looking myometrium between the uterus and the urinary bladder. A placenta is now seen, posteriorly compressing the endometrium.

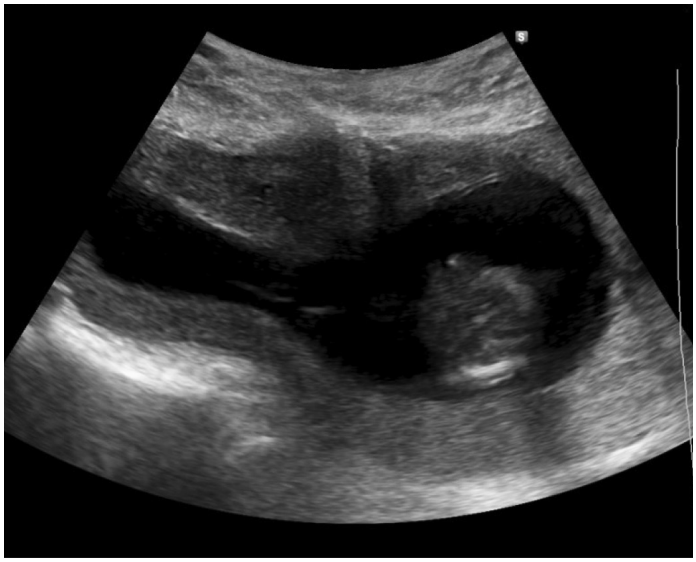

Figure 7. Transabdominal scan a 17 weeks and 1 day showing expansion of only the lower uterine segment

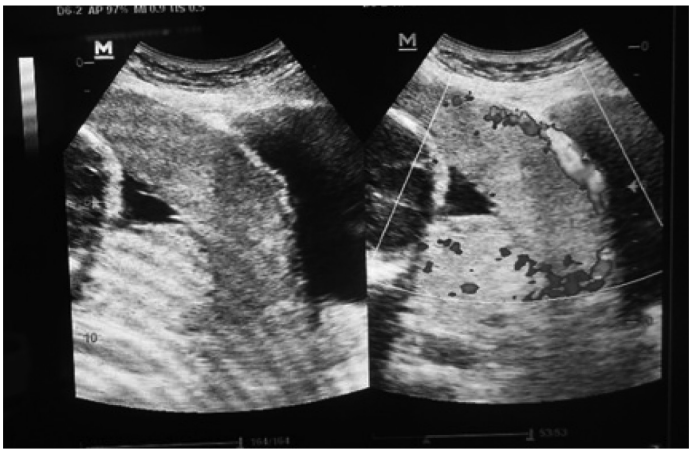

Figure 8. Transabdominal scan at 26 weeks showing a placenta totally covering the os and with sonographic findings suggestive of placenta accreta, possible percreta.

Third trimester

Subsequent ultrasound findings were consistent with placenta previa with accreta and possible percreta. (Figure 9). The pregnancy was allowed to continue without complications. She was admitted at 34 weeks for close monitoring and fetal surveillance.

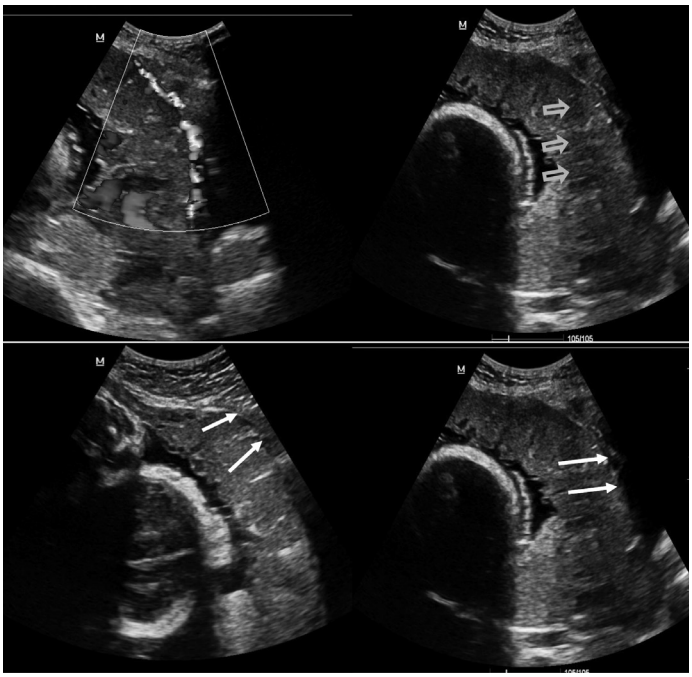

Figure 9. Gray-scale and color Doppler images showing sonographic diagnostic criteria for morbidly adherent placenta.
Top left: Hypervascularity in posterior bladder wall
Top right: Presence of placental lacunae
Bottom left: Loss/irregularity of echolucent area located between uterus and placenta (arrows).
Bottom right: Bladder line: thinning or interruption of hyperechoic interface between uterine serosa and bladder wall (arrows).

At 37 weeks, she underwent diagnostic cystourethroscopy, primary classical cesarean section, adhesiolysis, total hysterectomy with bilateral salpingectomy, cystorrhaphy, methylene blue instillation, omental flap interposition under spinal anesthesia. She delivered a live baby boy, 2600 grams, 37 weeks by pediatric aging with APGAR score of 9 remaining 9. Cut section of the uterus revealed a placenta measuring 14 x 12 x 3 cm implanted anteriorly from the midcorpus up to the posterior myometrium 3 cm from the cervical os with full thickness myometrial invasion at the lower uterine segment (Figure 15). Final histopathology report confirmed the diagnosis of placenta accrete (Figure 16).

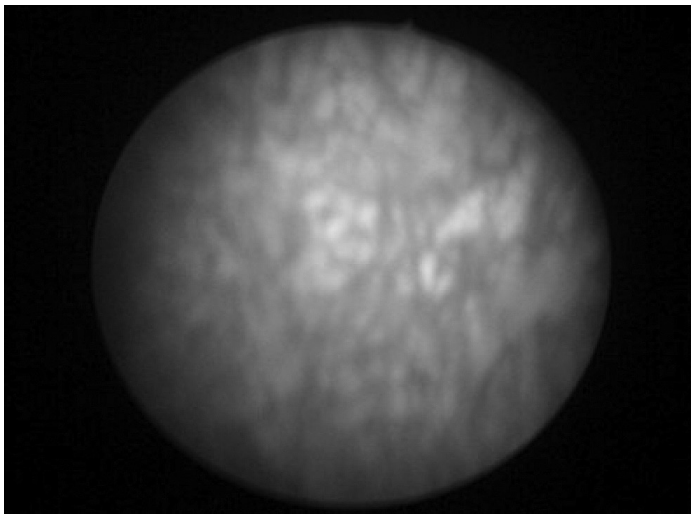

Figure 12. Hypervascularity in posterior bladder wall viewed through cystourethroscope.

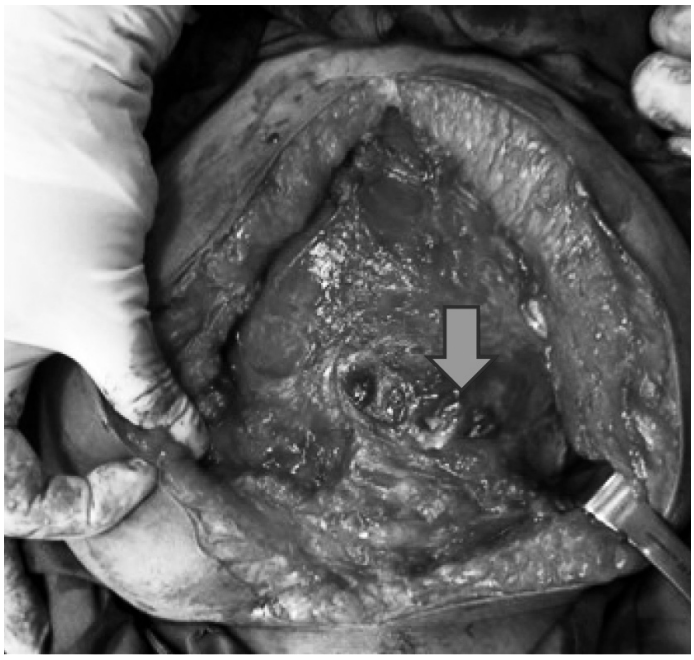

Figure 13. The bladder is densely adherent to the lower uterine segment**.**

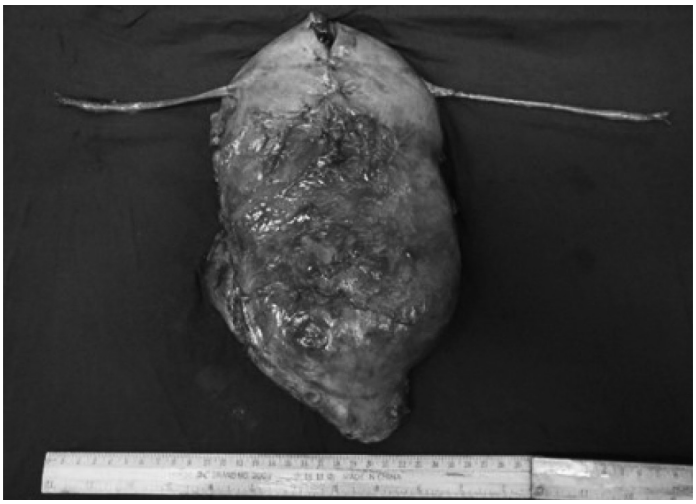

Figure 14. The placenta is implanted anteriorly from the midcorpus up to the posterior myometrium 3 cm from the cervical os with full thickness myometrial invasion at the lower uterine segment.

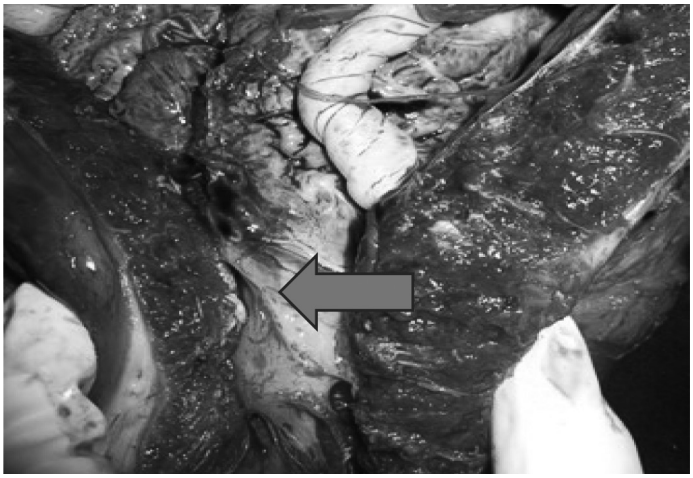

Figure 15. There is thinning and loss of myometrium with but with intact serosa. The thinnest portion of the serosa measures 0.1 cm

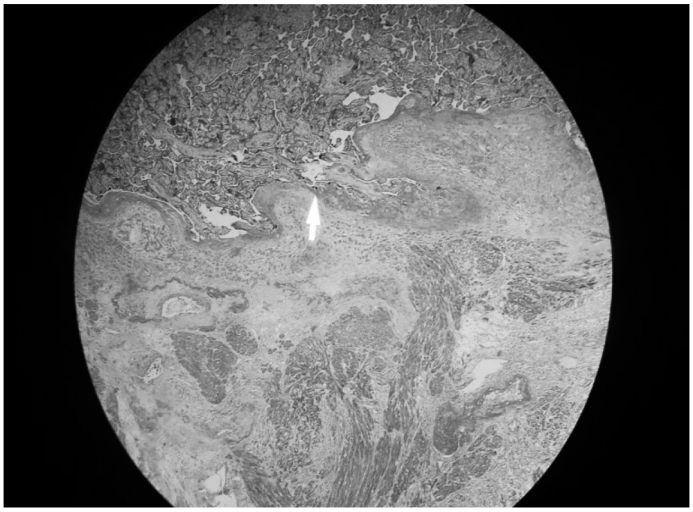

Figure 16. Histopathologic image of uterine tissue showing chorionic villi invading myometrium and absence of decidua basali

DISCUSSION

Cesarean scar pregnancy (CSP) is a rare form of ectopic pregnancy characterized by implantation of the fertilized ovum in the myometrium of the lower uterine segment, on the previous cesarean section scar.[3] The first case of CSP is reported by Larsen and Solomon in 1978.[4] The exact incidence is unknown but most literatures estimated it from 1/1800- 1/2500 of all cesarean deliveries.[4] The rate is 0.15% in women with a previous cesarean section and a rate of 6.1% of all ectopic pregnancies in women who had at least one cesarean delivery.[5] In Philippine General Hospital, there are only 2 cases of cesarean pregnancy reported for the year 2014 with local incidence of 1:2447.

The exact cause and mechanism of CSP is not well understood. Of all the theories proposed, the most acceptable is that the blastocyst enters into the myometrium through a microscopic dehiscent tract, which was created during the previous cesarean section, gynecologic surgery or even following removal of placenta.[1] This explains why the gestational sac in CSP is completely embedded in the myometrium, surrounded by fibrous scar tissue and separate from the endometrial cavity. There are also reports of intramural implantation secondary toin vitro fertilization (IVF) and embryo transfer. Other risk factors associated with CSP, although with limited studies to support its correlation, are the surgical technique used for uterine closure (single-layer, various suture material), the use of an intrauterine device, and previous history of pelvic inflammatory disease.[1]

Vial et al (2000), proposed two types of ectopic CSP. The first type has tendency to progress towards the cervicoisthmic canal or endometrial cavity and has the possibility to be carried to a viable pregnancy. This type is associated with life threatening massive hemorrhage from the implantation site. The second type involves deep implantation in the cesarean scar and involves growth of the gestational tissue towards the bladder and into the abdominal cavity. This type is associated with increased maternal morbidity and mortality secondary to uterine rupture, hysterectomy and massive hemorrhage.[6] The index patient has a type 1 CSP, hence was managed expectantly and was able to deliver to a term baby.

As with other ectopic pregnancies, early diagnosis is important in CSP because it allows for more treatment options and avoids the dangers of hemorrhage and uterine rupture.

Transvaginal ultrasound is the first line diagnostic tool for CSP, with a reported diagnostic sensitivity of 86.4%. Transvaginal ultrasound in combination with Color Flow Doppler further provides higher diagnostic accuracy with very few false positives. Doppler can show a distinct circular peritrophoblastic perfusion surrounding the gestational sac that can help delineate the CSP sac and location of the placenta in relation to the scar and proximity to the bladder. 3D-Ultrasound combined with the multiplanar views and surface-rendered images helps identify subtle anatomical details of a well-developed trophoblastic shell around the GS.[7]

The diagnosis of CSP is relatively easy if transvaginal ultrasound was done during early weeks of pregnancy because the small gestational sac and the implantation site can be identified accurately. Distinguishing between a CSP, cervical pregnancy, a lowly implanted intrauterine pregnancy, spontaneous miscarriage in progress and an intrauterine pregnancy with placenta accreta can be challenging. In the study done by Timor-Tritsch et al (2012), they mentioned that out of 751 cases of CSP identified, about 13.6% (107/751) has been misdiagnosed as cervical pregnancies, spontaneous abortion in progress (on its way to expulsion), or low intrauterine pregnancies. Therefore, reliable diagnostic criteria are required to establish the diagnosis.

For cervical pregnancy, the fetus and placenta implant in the cervix. Cervical pregnancy is diagnosed sonographically according to the following criteria: (1) presence of gestational sac or placental tissue dominantly within the cervix, (2) no evidence of intrauterine pregnancy, (3) visualization of an endometrial stripe (except in case of heterotopic pregnancy), (4) hourglass uterine shape with ballooned cervical canal, and (5) gestational sac with active cardiac motion below the internal os for viable pregnancy.[7] This was ruled out because the gestational sac was seen at the level of uterine isthmus and there was no ultrasonographic evidence of trophoblastic invasion of the cervix. Unlike CSP, in cervical pregnancy, there is a healthy (thick) myometrium intervening between the urinary bladder and gestational sac together with the ballooned cervical canal that give the uterus an hour glass appearance. In addition, cervical pregnancy is more likely to occur in women with no history of cesarean delivery and rarely could progress to a term pregnancy as seen in this case.

Color Doppler scanning is also useful in differentiating between the spontaneous miscarriage in progress and cervical pregnancy. In cervical pregnancy, Doppler studies show characteristic patterns of trophoblast with high flow velocity and low impedance, while in abortion in progress the gestational sac will be mobile with no Doppler evidence of blood flow.[8] To further identify a cervical stage of abortion rather than a cervical pregnancy, “sliding sign” can be ascertained by ultrasound. If the gestational sac slides with gentle pressure on the cervix with the ultrasound probe, a cervical abortion is often considered.

In contrast with CSP, intrauterine pregnancy in the process of abortion rarely would have an embryo with a

cardiac activity. Also, the internal os in cervical pregnancy and abortion maybe open or close but is always close in CSP.

CSP should also be distinctly differentiated from the intrauterine pregnancies with placenta accreta. In the latter condition, the gestational sac is implanted within the endometrial cavity but there is abnormal invasion of the placenta to the myometrium, while a CSP is an ectopic pregnancy that is implanted entirely at the previous cesarean scar and is surrounded by myometrium and fibrotic tissue.[9]

CSP is suspected by ultrasound when the uterine cavity and cervical canal are completely empty and the GS develops in the anterior part of the uterine isthmus, and there is no myometrium between the bladder and sac. The diagnosis is usually made by ultrasound but hysteroscopy, laparoscopy or even MRI can also be helpful [(10)]. In this case there was conflicting results in the distinction between previous CS pregnancy and a lowly implanted intrauterine pregnancy on ultrasound. MRI can accurately detect the exact location of pregnancy, thus confirming the diagnosis of CSP in this case.

Timor-Tristsh et al (2012), suggested the following criteria to diagnose CSP using transvaginal ultrasound: (1) visualization of an empty uterine cavity as well as an empty endocervical canal, (2) detection of placenta and/or a GS embedded in the hysterotomy scar, (3) in early gestation ,</ 8 weeks), a triangular GS that fills the niche of the sac; at >/8 weeks this shape may become rounded or even oval, (4) a thin (1-3 mm) or absent myometrial layer between the GS and the bladder, (5) a closed and empty cervical canal, (6) the presence of embryonic/fetal pole and/or yolk sac with or without heart activity, and (7) the presence of a prominent and at times rich vascular pattern at or in the area of CS scar in the presence of a positive pregnancy test. All of these criteria had to be present to diagnose CSP. In the index case, all of these criteria were identified.

The most common presenting symptom of CSP is painless vaginal bleeding as seen in this case. This is supported by the study conducted by Michener and Dickinson in 2009, 9 out of 13 patients manifested with painless vaginal bleeding. The median gestation at diagnosis was 6.8 weeks. Meanwhile, in the study made by Silver et al, they reported that 37% of 57 women were symptomatic, 30% had painless vaginal bleeding, 16% had painful vaginal bleeding and 9% experienced abdominal pain without vaginal bleeding. Because of these, many cases of CSP go undetected initially and only get recognized once the uterus has ruptured and they are in hypovolemic shock. Thus, early and accurate diagnosis of the condition is crucial because a delay in diagnosis may lead to life-threatening complications. Close monitoring should be rendered when the diagnosis is suggested by ultrasound.

In this case, the diagnosis of CSP by ultrasound and MRI was established. The mutual decision was to avoid any intervention and to manage this pregnancy expectantly. Repeat ultrasound after 2 weeks (Figure 4) showed that the placenta was growing towards the uterine cavity. The possibility of hemorrhagic complications such as uterine rupture and massive hemorrhage leading to maternal mortality and morbidity were reiterated to the patient. Also, the possibility of developing into placenta accreta/percreta was well-explained to the patient.

At 26 weeks (Figure 8), repeat scan showed that the placenta is totally covering the os and with sonographic findings suggestive of placenta accreta, possible percreta such as loss of retroplacental hypoechoic zone, progressive thinning of the retroplacental hypoechoic zone, presence of multiple placental lakes, thinning of the uterine serosa-bladder wall complex and elevation of the tissue beyond the uterine serosa. Color Doppler evaluation further supported the diagnosis with findings of dilated vascular channels with diffuse lacunar flow, hypervascularity linking placenta to bladder.

The evolution of a CSP into placenta accreta/percreta at term is a common complication once pregnancy is allowed to continue. It was reported in the study of Nagi et al (2007), that the incidence of abnormally adherent placentas is 40% when the placenta is implanted over the uterine scar. It has also been shown that abnormally adherent placentas account for 50-65% of all obstetric hysterectomies. In our index patient, intraoperatively, the placenta was firmly adherent to the myometrium but without evidence of bladder invasion. An emergency hysterectomy was performed. Final histopathology report confirmed the diagnosis of placenta accreta.

Once the diagnosis of CSP has been established, the patient should be counseled about her options. In general, the treatment should be individualized, based on the patient's age, number of previous cesarean deliveries, number of children and the expertise of the clinicians managing her case. Options include termination of the pregnancy or continuation of the pregnancy with the possibility of delivering a live offspring. The patient should be informed that a morbidly adherent placenta may occur and often necessitates emergency hysterectomy.

There are no universal treatment guidelines for the management of CSP, especially in more advanced pregnancies. Reported management include expectant management, systemic methotrexate therapy, local injection of methotrexate, gestational sac aspiration, dilatation and curettage, surgical laparotomy/hysterotomy, hysteroscopy, laparoscopy.[8]

Expectant management of a viable pregnancy, once the diagnosis is certain, is understandable but there are

risks of life threatening complications such as uterine rupture which is very common as CSP progresses. There is also a danger of invasion of the bladder by the growing placenta. Sadeghi et al (2010) reviewed 268 cases from various case reports and series showed that out of twenty-one (21) cases which were managed expectantly, fourteen (14) women or 67% required subsequent intervention, and six of these cases (29%) ultimately required hysterectomy. There have been a few cases of CSP that resulted in viable infants. Usually, these are pregnancies where the gestational sac grows towards the uterus and eventually becoming mostly intrauterine pregnancy, like what happened to the index case. According to Ioannis et al (2008), expectant management can also end into an spontaneous first trimester miscarriage in about 44% of cases.

In the study of Timor-Tritsh (2014), in those patients that expectantly managed, 9 out 10 patients were able to deliver live babies between 32-37 weeks age of gestation. All patients underwent cesarean section hysterectomy and with a histopathologic diagnosis of placenta percreta. Hence, they concluded that expectantly managed CSPs during the first trimester could evolved into pregnancies with morbidly adherent placenta.

The therapeutic approach of cesarean scar pregnancy is still considered to be a dilemma. No one can guarantee uterine integrity because the number of cases in the literature is still small. But it seems that the earlier the diagnosis, the more minimal the therapeutic approach.

Most clinicians believe that the only reasonable treatment for CS pregnancy is to end the pregnancy but the best method is still being debated. Termination of pregnancy was initially offered to the patient but she opted for an expectant management. ■

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