Cesarean scar pregnancy "A rarity no more?" A report of 2 cases*

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ABSTRACT

Cesarean scar pregnancy is the rarest form of ectopic pregnancy. In the Philippines, little is known about its incidence and occurrence. However, increasing rates has been documented worldwide, closely related to the increasing cesarean section rates. This paper reports two cases of cesarean scar pregnancy who both presented with vaginal bleeding. The first case, a Gravida 6 Para 5 (5005), while the second case, a Gravida 3 Para 2 (2002). Both diagnosed early by ultrasonography but managed differently. The first case, managed by hysterectomy, while the second case, managed conservatively by laparoscopic excision of the cesarean scar pregnancy. This paper intends to raise awareness of the increasing incidence of cesarean scar pregnancy, its pathophysiology, different options in the diagnosis and management.

Prevention is the key to decrease the incidence of cesarean scar pregnancy. To achieve this, reducing the cesarean section rate should be the primary goal.

Keywords: Ectopic pregnancy, Cesarean scar pregnancy (CSP), Cesarean section

INTRODUCTION

Ectopic pregnancies comprise about 1 to 2 % of all pregnancies. Among all types of which, cesarean scar pregnancy is the rarest form accounting for 6.1 % of all ectopic pregnancies with an incidence rate of approximately 1:1800 to 1:2216.[1] Because of its rarity, there are still no evidence based standards of practice for its diagnosis and management.

With delayed detection, it may lead to massive hemorrhage, uterine rupture and other life threatening complications.

In the Philippines, little is known about its incidence and natural history. Statistics from Philippines Obstetrics and Gynecology Society reported about 9,365 cases of ectopic pregnancies within the last 5 years (2010-2016). However, no report of CSP was ever recorded. This paper intends to promote awareness and develop accurate reporting of cases of CSP in the Philippines, as it's increasing rate worldwide pose great concern due to its life threatening outcomes.

OBJECTIVES

General Objectives: To discuss 2 cases of cesarean scar pregnancy diagnosed early by ultrasound imaging and managed differently.

Specific Objectives:

1. To review the factors involved in the increasing incidence of cesarean section scar pregnancy (CSP)
2. To discuss the pathophysiology of the disease
3. To discuss the different options in diagnosis and management of cesarean section scar pregnancy
4. To make recommendations on how to decrease the incidence of cesarean scar pregnancy

CASE REPORTS

First case, a 37 year-old, G6P5 (5-0-0-5), 7 6/7 weeks, admitted due to vaginal bleeding. Pregnancy test was positive. Two days prior to admission, patient experienced profuse vaginal bleeding, hypogastric pain, vomiting, body malaise and dizziness. The persistence of the symptoms prompted consult at our institution. Transvaginal ultrasound revealed a live fetus, compatible to 9 weeks and 1 day age of gestation, measuring 2.9 x 2.3 cm located at the previous CS scar at the anterior isthmic portion without subchorionic hemorrhage (Fig.1).

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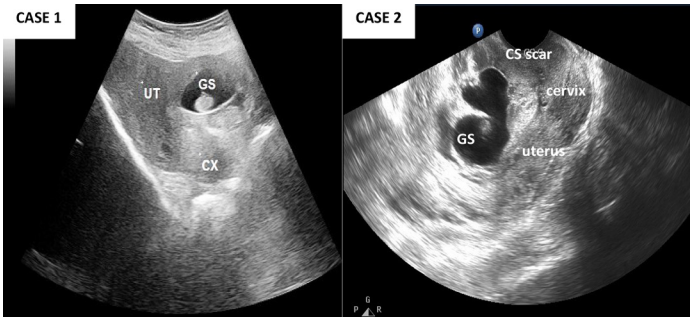

Figure 1. Endocervical canal and uterus is empty. Gestational sac with an embryo and a yolk sac is seemingly located within the cesarean scar anterior to the uterus.

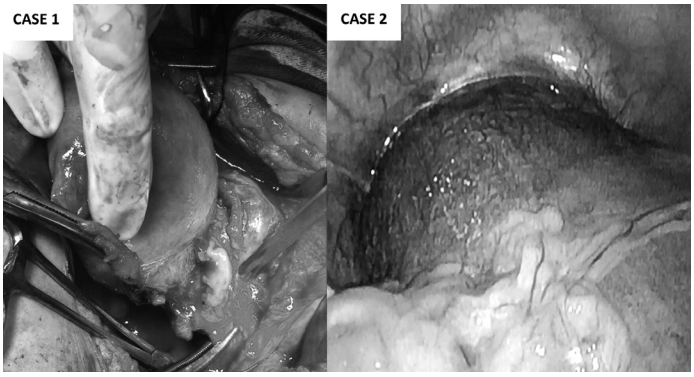

Figure 2. Case 1: Conceptus noted to be implanted anterior to the myometrium, with a thin membrane separating from the urinary bladder. Case 2: A highly vascular mass at the serosal surface was noted approximately measuring 3.0 x 2.0 x 6.0 cm.

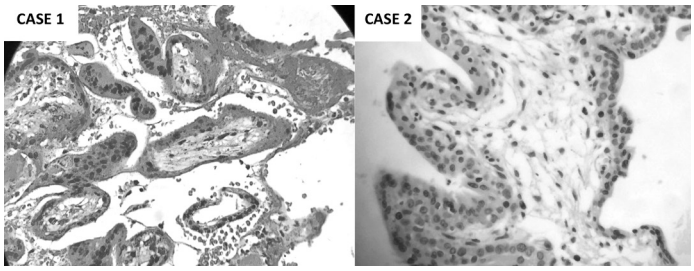

Figure 3. Poorly vascularized chorionic villi representing immature placental tissues

First and second pregnancies were delivered by normal delivery. Her third was delivered via low segment cesarean section due to fetopelvic disproportion. Subsequent pregnancies were delivered by repeat cesarean section.

On physical examination, patient was ambulatory, with stable vital signs. Abdomen has previous midline scar, flat, soft, no mass, non-tender. On speculum examination, cervix is clean looking with minimal bleeding per os. Internal examination revealed closed cervix, no cervical motion tenderness, uterus slightly enlarged, no adnexal mass nor tenderness. Diagnosis was G6P5(5-0-0-5), Cesarean Section Scar pregnancy, 9 weeks, Unruptured. She was scheduled for total abdominal hysterectomy.

On laparotomy, there was no hemoperitoneum, uterus slightly enlarged with numerous varicosities at the isthmic portion. A bulging mass measuring 5.0 x 4.0 cm was at the previous CS scar and adherent to the bladder. After ligation of the round, uteroovarian, and uterotubal ligaments; the vesicouterine peritoneum was dissected downwards separating the bladder from the uterine wall. The products of conception were expelled at its implantation site (Fig.2). Completion of the hysterectomy procedures were performed afterwards. A small rent was seen at the bladder upon methylene blue instillatio. Cystorrhaphy by a urogynecologist ensued. Estimated blood loss was 1,100 cc corrected by blood transfusion. Patient was discharged improved with indwelling foley catheter. Histopathologic examination confirmed a cesarean scar pregnancy (Fig.3).

One week post-operation, she complained of urinary leakage. Voiding cystogram confirmed presence of vesicovaginal fistula. Catheter was retained for one month and removed per patient's request. She has been asymptomatic ever since.

The second case, a 33 year-old, G3P2 (2-0-0-2), 7 5/7 weeks, admitted due to vaginal spotting.

Three weeks prior to admission, patient experienced vaginal spotting. Self-pregnancy test was positive. Few hours prior, she experienced low back pain and consulted at a hospital where transvaginal ultrasound revealed a live pregnancy, 8 weeks, with a gestational sac noted at the lower uterine segment. Suspicious of a cesarean scar pregnancy, she was transferred at our institution.

Her first pregnancy was delivered by low segment cesarean section due to CPD. Second pregnancy was delivered by repeat CS last 2015.

On physical examination, vital signs were stable, abdomen has a pfannenstiel scar, no mass noted and non-tender. On pelvic examination, a clean looking cervix with minimal bleeding per os was noted. cervix closed with no motion tenderness, uterus slightly enlarged, no adnexal mass nor tenderness. Repeat transvaginal sonography revealed a live pregnancy at 8 weeks and 3 days with an irregularly shaped gestational sac within is an embryo and yolk sac measuring 6 x 5 cm implanted at the anterior lower uterine segment, probably in the previous cesarean scar with subchorionic hemorrhage (Fig.1). Diagnosis was T/C Cesarean Scar pregnancy, 7 weeks and 5 days, unruptured. With desire for future pregnancy, she was referred to a reproductive endocrinologist and infertility specialist for diagnostic laparoscopy. Serum beta-hCG was elevated at 198, 289.18 mIU/ml.

On laparoscopy, there was no hemoperitoneum, uterus was enlarged to 12 weeks' size with a 3.0 x 2.0 x 6.0 cm highly vascularized mass protruding at the serosal surface of the lower uterine segment (Fig.2). Upon dissection of the uterovesical peritoneum, the products of conception were expelled and evacuated. Hemostasis was

achieved. Estimated blood loss was 1,400 cc. CBC revealed severe anemia with hemoglobin count of 63 g/L which was improved by three units packed RBC to a hemoglobin count of 84 g/L. Patient was discharged at postoperative day 3. Histopathologic result confirms cesarean scar pregnancy (Fig.3).

On her follow up, she complained of vaginal spotting. Speculum examination revealed a clean looking cervix with minimal bleeding per os, internal examination was unremarkable. Repeat transvaginal ultrasound showed an irregular cystic structure with strong vascular flow seemingly encroaching the endometrium within the lower uterine segment. Impression was gestational trophoblastic disease versus arteriovenous malformation. Weekly B-HCG monitoring was done. Decreasing values were noted and was normal at 5th week post-operation with resolution of the vaginal bleeding (Fig.4). After one month, repeat ultrasound was unremarkable, with no niche noted.

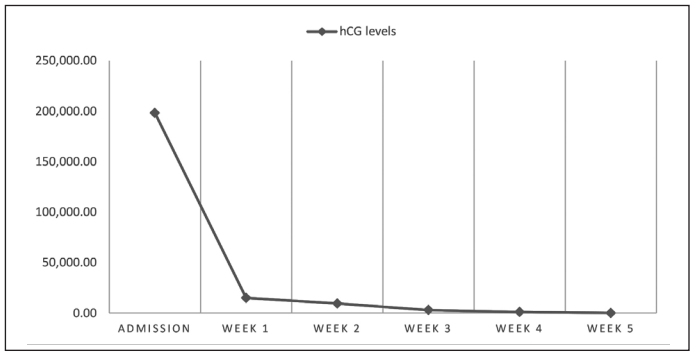

Figure 4. Serial hCG Monitoring

CASE DISCUSSION

Ectopic pregnancy comprises 1% to 2% of all pregnancies and the leading cause of pregnancy-related deaths during the first trimester, accounting for 10% of all maternal deaths.[2]

Cesarean scar pregnancy (CSP) is the rarest form of ectopic pregnancy, accounting for 6.1% of all ectopic pregnancies in women who had at least one cesarean delivery and a rate of 0.15% in women with previous CS regardless of the number of cesarean sections done.

In 2002, Fylstra et al, only found 19 cases of CSP reported in the English medical literature since 1966. Furthermore, in 2004, the same authors searched for case reports that were made after 2002, and found 66 new cases in a span of two years. Then recently in 2012, a survey of the literature found 751 case reports. From these statistics, it can be concluded that the incidence is rising.[3]

What could have caused the increasing incidence of CSP? Recently, several studies have emphasized that the rise of CSPs worldwide coincides with the increasing rates of cesarean section. Potentially making them a dangerous combination. Over the past three decades, rates of CS have been increasing despite the ideal rate of 10-15% set by the World Health Organization (WHO) in 1985. Despite efforts made by the WHO and other world administrations, the CS rates have been noted to rise steeply. In the UK, their CS rates increased from 12 to 29% between 1990 and 2008 as documented by Betran et al. In 2011, the USA reported one in three women delivered by CS. Based on the CS rates of China, it was reported to have risen by 2% in 1985 to 36 to 58% in 2010. In Brazil, from 15% in 1970 it increased to 80% in the private sector in 2004.[4]

Even in the Philippines, the 5-year statistics of CS rates taken from Philippine Obstetrics and Gynecology Society of the Philippines (POGS) showed an increasing trend from 2011 and reached its peak by the year 2014 at 38%. By the year 2015, it was reduced to at least 31%, but still comprises about one third of all deliveries made that year and above the rates set by WHO. The primary CS rates in our institution also showed an increasing trend as well. It had a steady rate at 23 to 25% from year 2012 to 2014 proceeded by a remarkable decline on 2015 at 17.07%. Even so, from the year 2015 to 2016 it showed an extensive rise to 26.36%, the highest recorded primary CS rate at our institution. Primary CS cases were probably delivered via repeat CS. Hence, the likelihood of CSP and its consequences. This reasoning will further support the observation that the rise in CSP can be attributed to the increasing CS rates worldwide.

The inflation of CS rates has been attributed to several factors including cesarean delivery on request, increased prevalence of high risk pregnancies such as advanced maternal age, subjective indications like non-reassuring fetal status and arrest in cervical dilatation. On the other hand, a decreased number of Vaginal Birth after Cesarean Section (VBAC) and vaginal breech delivery were observed.

Another contributory factor are changes in operative techniques of closing the uterus through a single layer versus double or multiple layer, a common practice from the past. Single layer technique is associated with the development of a phenomenon known as the 'niche'. A hypoechoic area within the myometrium of the lower uterine segment, reflecting a discontinuation of the myometrium at the previous CS site, providing a ground for the implantation of a conceptus.[6]

Yazicioglu et al. (2006) made a significant study among 98 patients where he used two different techniques in closing the uterus. One technique is closing the single layer full-thickness of the uterus including the endometrial layer and the other is multiple layer split-thickness closing wherein the endometrium was excluded. The results revealed lower rates of formation of cesarean scar defect

among those who underwent multiple layer closure. Single layer closure cannot guarantee accurate alignment of the uterine edges, increasing the formation of a CS defect.[7]

In both patients, there was no previous ultrasound prior to pregnancy to note for any presence of "niche" and the manner of uterine closure during the previous CS were unknown.

Cesarean scar pregnancy is the invasion of the conceptus on the myometrium of a previous CS scar. The most plausible theory suggests that the blastocyst enters into the wall through a microscopic dehiscent tract, created by a trauma that occurred in association with a cesarean section or any uterine surgery such as dilatation and curettage (Cheng et al., 2003), or following manual removal of the placenta (Fylstra, 2002). In Vitro Fertilization could also represent a rare mechanism (Seow et al., 2000, 2004), or can even occur in the absence of any previous uterine surgery (Hamilton et al., 1992).[8]

The most common manifestation is painless vaginal bleeding but presenting symptoms of CSP aren't specific. In a study by Silver et al., presenting symptoms of CSP were observed in 57 pregnant women: 37% are asymptomatic, 38% have painless vaginal bleeding, 16% had painful vaginal bleeding, and 9% experienced abdominal pain without vaginal bleeding.[9] *In these two cases, both presented with vaginal bleeding. The first case had hypogastric pain while the second case presented with low back pain, representing a myriad of its symptoms, in which, at an early pregnancy, can easily be mistaken as abortion.*

Diagnosis requires a high index of suspicion, especially, on ultrasonography, no intrauterine gestational sac can be identified. Usual differential diagnoses for CSP is cervical ectopic pregnancy, placenta accreta and abortion in progress or incomplete abortion.

The gold standard for the diagnosis is transvaginal ultrasonography (TVUS) with an accuracy rate of 89%. It is based on finding a gestational sac at the site of the previous CS scar with an empty uterine cavity, cervix, and strong color flow on the anterior myometrium, as well as a thin myometrium adjacent to the bladder.[10] These criteria may rule out the differential diagnoses mentioned above. *Both of our cases, fulfilled such criteria.*

Management of CSP still remains controversial because of the lack of evidence based standards for its treatment. Management options cater to individual patients depending on gestational age, fetal viability, severity of symptoms, serum hCG levels and ultrasonography findings. Options may vary, it can be conservative, either medical or surgical approach, if the woman is still desirous of pregnancy. It can also be radical involving surgical removal of the uterus for those who have completed their childhood bearing career.

In the medical approach, several agents can be used, one of which is Methotrexate. This can be used in fulfillment of the following criteria: unruptured CSP, size <3.5cm, no cardiac activity and hCG levels <5,000 mIU/ml. Administration may be systemically or locally by injecting directly to the sac along with potassium chloride into the fetal heart. However, ethical consideration is an issue. Other nonsurgical approach can be through uterine artery embolization (UAE). Undoubtedly, the nonsurgical interventions are less invasive and has lesser complications. Conversely, the downsides of non-surgical management entails longer time frame for follow up until normalization of hCG levels and serial ultrasonographic monitoring for the shrinkage of the sac.

Surgical interventions, usually laparotomy and resection of ectopic sac along with previous scar tissue is done. With skilled hands, laparoscopic excision alone would suffice. Serial hCG monitoring is usually done with conservative management. This is due to the anticipated slow decline in the hCG levels in CSP since the gestational sac is implanted on a fibrous tissue. *As with the second case, the patient is still desirous of future pregnancies, hence, a conservative approach was done. Because of the size of the gestational sac, as well as the presence of cardiac activity, Methotrexate was not an option.* Recurrence of CSP is still possible in the subsequent pregnancies thus an earlier surveillance is required to rule it out. *In our second case, serial ultrasound and weekly hCG monitoring were done for post-surgical surveillance. By five weeks' post-operation, hCG was normal and ultrasound was unremarkable.*

According to Dr. Timor-Tritsch in 2012, increases in hCG concentrations could be expected with treatment. Secondary treatments like embolization and even hysterectomy were considered not because of bleeding but due to observation of post treatment rise in hCG cycle. With the knowledge of the naturally occurring increase in the hCG volume within the blood vessels, secondary resolution could be avoided. *As with the second case, her hCG levels remained high few weeks postoperatively but gradually declined to normal levels by 5th week post-operation. Through observation and knowledge of the natural pattern of the hCG level in CSP, over treatment can be avoided.*

In contrast, for patients who are not desirous for future pregnancies, radical surgery can be done such as hysterectomy. *In the first case, the patient is multigravida, ergo, total abdominal hysterectomy was the optimal choice. It gives the best access to the pelvic structures, controlled operative field including ligation of bleeders and lesser operative time.* Such radical procedures will also allow complete removal of the products of conception. Hence, further monitoring will not be any more warranted.

SUMMARY

In summary, this is a report of two cases of cesarean scar pregnancy.

Both presented with vaginal bleeding and diagnosed promptly through sonography. The first case is not desirous of pregnancy; hence hysterectomy was performed. On the other hand, the second case has future fertility plans, therefore, conservative laparoscopic excision of the conceptus was done.

A growing incidence of CSP was reported which coincides with the increasing rates of CS. More cases of CSP are being recognized at present with the advent of imaging technology such as ultrasonography. The practice of a single-layer suturing in CS may also contribute to its increasing occurrence. Logically, the blastocyst enters into the wall through a dehiscent tract from a trauma created by uterine procedures. Most common manifestation is painless vaginal bleeding but symptoms may vary. Transvaginal sonography is the gold standard for diagnosis. Management which includes treatment and follow-up depends on the age, fertility desires, gestational age, fetal viability, size of the mass and hCG levels. Management maybe through methotrexate injection, embolization, excision by laparoscopy or laparotomy in order to preserve the uterus. This approach entails monitoring with serial hCG determination and sonography. For those who has completed their childbearing capacity, hysterectomy may be the optimal management.

RECOMMENDATIONS

The following are the recommendations this paper intends to impart on how to decrease the incidence of CSP:

1. Strict adherence to the recommended indications for performing cesarean sections
2. Trial of labor or VBAC for patients who fulfill the criteria
3. Breech vaginal delivery for multiparous women
4. Closing of the uterus in multiple layers to lessen niche formation
5. Accurate reporting on types of ectopic pregnancies for detailed statistics of CSP in the Philippines

Cesarean scar pregnancy, a previously rare entity, now occurring more than ever, should be recognized, reported and most importantly prevented. ■

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