Outcome of the Expectant Management of Ten Cases of Cesarean Scar Pregnancy in Patients Who Refused Termination of Pregnancy

Atif Fazari 1, 2, Parveen Bhanu Mohammed 3, Asma Fahad 4

1. Faculty of Medicine, University of Medical Sciences & Technology, Khartoum, SDN 2. Obstetrics & Gynecology, Latifa Hospital, Dubai Academic Health Corporation, Dubai, ARE 3. Obstetrics, Latifa Hospital, Dubai Academic Health Corporation, Dubai, ARE 4. Obstetrics and Gynaecology, Latifa Hospital, Dubai Academic Health corporation, Dubai, ARE

Corresponding author: Asma Fahad, asma.ahmad2011@gmail.com

Abstract

Background:

Expectant management of Cesarean scar pregnancy (CSP), in patients who refused termination of pregnancy, and continued as placenta accreta spectrum (PAS), is possible with multidisciplinary care with careful monitoring in tertiary center having expertise and excellence in the management of placenta accreta involving highly accurate ultrasound monitoring as a tool in the prenatal diagnosis and further monitoring and management, which enable appropriate surgical strategies and accurate techniques achieving best maternal and fetal outcomes with minimal blood loss without major maternal mortality and morbidity. This study is aimed to evaluate expectant management in those patients.

Methods:

This is a retrospective study including 10 patients with previous history of uterine scar, with CSP diagnosed in first trimester who refused to terminate their pregnancy and continued as PAS were studied during the period of 4 years, from 2018 to 2022, who were managed in our hospital. The main outcomes were analyzed depending on factors like the gestational age at diagnosis, CSP type (endogenous or exogenous), gravida, parity, previous cesarean section history, gestational weeks at termination, amount of blood loss, duration of surgery and correlating the USS findings with surgical findings and to assess obstetric and neonatal outcomes and complications in expert hands.

Results:

Out of 10 cases, 9 cases were delivered in 3rd trimester [around 34 weeks’ gestation]. 7 cases were elective and 3 cases were done as emergency surgery. 4 cases were exogenous and 6 cases identified as endogenous type, at diagnosis during early gestation. 7 cases had cesarean hysterectomy, and 3 of them had uterine wall reconstruction surgery (with focal accreta). 4 cases needed blood transfusion. Average duration of surgery took around 2.5 to 5 hours. There were no miscarriages, no maternal and neonatal deaths, and no significant obstetric complications like rupture of uterus, major obstetric hemorrhage.

Conclusion:

Even though CSP is potentially life threatening condition because of serious complications like placenta accreta if continued, however expectant management is possible under multidisciplinary team care, strictly adhering to clinical protocols and surgery with accurate techniques to reduce obstetric hemorrhage.

Introduction

Cesarean scar pregnancy (CSP), Is defined as implantation of pregnancy into the myometrial defect in the previous uterine scar or defect in the scar. CSP poses a diagnostic and management challenge. If unrecognized, left untreated or patient refused termination of pregnancy, which will continue as viable pregnancy, transform as morbidly adherent placenta [MAP] / Placenta accreta spectrum, associated with risk of major obstetric hemorrhage, peri-partum hysterectomy and maternal mortality and morbidity and both conditions share same pathology and histology. [1,2]

There are mainly two types of CSP:

Type 1 CSP [ Endogenous]: Pregnancy is completely located in the uterine cavity, and continues to grow inside the uterine cavity and deeply into the myometrium, but does not bulge out of the serosa, this type of CSP, has more potential to reach viable gestation [till 3rd trimester] as placenta accreta or increta and associated complications. [1,3]

Type 2 CSP [ Exogenous]: Pregnancy almost completely located in the lower uterine segment, and bulges out of the serosa and grows out towards the bladder, as placenta percreta, and cause potential complications like
scar rupture and intraperitoneal bleeding and significant mortality and morbidity. [1,3]

Type 3 CSP, [Mixed]; has characteristics of both type 1 and type 2, which is very rare.

Ultrasound (Trans vaginal) is highly accurate in the prenatal diagnosis, monitoring and management of CSP evolving to placenta accreta spectrum [PAS]. Ultrasound findings can be highly indicative of the depths of villous invasion, vascularity and can enable surgical strategies and predict clinical outcomes. [4]

These are some characteristic ultrasound findings of PAS :
1) Loss of Retro placental hypoechoic clear Space.
2) Reduced Uterine Wall Thickness:
3) Myometrial thickness less than 3 mm
5) Increased vascularity is seen in any MAP/PAS:
6) Increased vascularity is seen in any MAP/PAS:

Myometrial thickness less than 3 mm
Increased vascularity is seen in any MAP/PAS:
Numerous large blood vessels are often seen surrounding the myometrium, Invasion can also create an irregular bladder wall with extensive associated vascularity. [5]
4) Dubais triad or Fazari's sign [stairs step appearance]:
5) "Equal sign" - Crossing /bridging vessels (defined as 2 parallel vesqels [neovascularization], depicted by color Doppler over the uterovesical junction and towards the bladder wall interconnecting bridging vessels perpendicular to both uterovesical walls. [4]
6) Bulging of urinary bladder wall.

Materials And Methods

This is a retrospective study including 10 patients with previous history of uterine scar, with cesarean scar pregnancy(CSP) diagnosed in first trimester who refused to terminate their pregnancy and continued as placenta accreta(MAP/PAS) were studied during the period of 4 years, from 2018 to 2022, who were managed in our hospital. After Diagnosis of CSP, patient will be followed in maternal-fetal medicine(MFM) unit with serial ultrasound scans (2D USS) along with Doppler sonography, the placenta with retro placental myometrium and bladder were mapped, when bladder is partially full, evaluate the progression of the placenta accreta spectrum(PAS), to observe and look for the above mentioned characteristic USS features of PAS. Meanwhile patient is counseled thoroughly regarding potential complications of major obstetric hemorrhage, blood transfusions and hysterectomy, take the consent and plan for the surgery i.e. classical cesarean section with cesarean hysterectomy at 34 to 35 weeks. Surgery was done in our hospital and during surgery, protocols are strictly followed by multidisciplinary team, involving 2 senior OBGYN consultants , senior anesthesiologist, including the presence of urologist [preoperative ureteric stenting was done to all cases to avoid urinary tract injuries] and vascular surgeon on standby and equipped blood bank services and intensive care unit. Midline longitudinal incision, classical cesarean section and proceed to cesarean hysterectomy, if placenta not get separated. Major artery ligation was also needed in some cases. Accurate surgical strategies and techniques, involving meticulous adhesiolysis, after introduction of neat dissection technique - Birds' picking seeds' technique [6] are applied as per the prenatal USS vascular mapping, to reduce obstetric hemorrhage. Results were analyzed depending on factors like the gestational age at diagnosis, CSP type, gravida, parity, previous cesarean section history, gestational weeks at termination, amount of blood loss, duration of surgery and correlating the USS findings with surgical findings and to assess obstetric and neonatal outcomes and complications in expert hands.

Results

Out of 10 cases, 9 cases were delivered in 3rd trimester [up to an average of 34 weeks' gestation], one case was delivered at 27 weeks' gestation, out of 9 cases in third trimester group, 2 cases delivered at 30 and 31 weeks of gestation. 7 cases underwent elective and 5 cases were done as emergency surgery. Four cases were exogenous (type 2 CSP) and 6 cases identified as endogenous type I CS, at diagnosis during early gestation. Seven cases had cesarean hysterectomy (7/10), and 3 of them had uterine wall reconstruction surgery (who had focal accreta). Four (4/10) patients needed blood transfusion [ on average of maximum 2 to 4 units] six of them did not needed blood transfusion. Average duration of surgery took around 2.5 to 5 hours, overall of around 5 hours. 4 patients were admitted to ICU. All 10 cases were diagnosed as placenta accreta pathologically and histologically, out of them 5 cases were found out to be focal accreta, where hysterectomy was not needed, in all cases there was accurate correlation between characteristic ultrasound features and surgical findings. There were no miscarriages, no maternal deaths, and no significant obstetric complications like rupture of uterus, major obstetric hemorrhage. There was no statistical difference in patients found, regarding the number of gravidity, parity and previous cesarean section. Considering Neonatal outcome (3/10) were admitted to NICU.
Summary of Results are shown in Table 1:

<table>
<thead>
<tr>
<th>Sl No</th>
<th>No.of LSCS</th>
<th>Type of CSP</th>
<th>GA at Diagnosis</th>
<th>Follow up scan</th>
<th>GA at Surgery</th>
<th>Intervention</th>
<th>Surgery</th>
<th>Blood Transfusion</th>
<th>Duration</th>
<th>Ultrasound findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>4</td>
<td>Exo</td>
<td>6, 1, 2, 3, 4, 5</td>
<td>30</td>
<td>Em</td>
<td>HYS</td>
<td>2</td>
<td>3 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>2</td>
<td>2</td>
<td>Exo</td>
<td>8, 1, 2, 3, 4, 5</td>
<td>33</td>
<td>El</td>
<td>HYS</td>
<td>0</td>
<td>4 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>3</td>
<td>4</td>
<td>Exo</td>
<td>7, 1, 2, 3, 4, 5</td>
<td>34</td>
<td>El</td>
<td>HYS</td>
<td>0</td>
<td>2.5 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>4</td>
<td>1</td>
<td>Exo</td>
<td>6, 1, 2, 3, 4, 5</td>
<td>33</td>
<td>El</td>
<td>HYS</td>
<td>0</td>
<td>4.5 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>5</td>
<td>3</td>
<td>Endo</td>
<td>9, 1, 2, 3, 4, 5</td>
<td>27</td>
<td>Em</td>
<td>HYS</td>
<td>4</td>
<td>3 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>6</td>
<td>1</td>
<td>Endo</td>
<td>8, 1, 2, 3, 4, 5</td>
<td>35</td>
<td>El</td>
<td>UWRS</td>
<td>2</td>
<td>5 hrs</td>
<td></td>
<td>Free UB wall</td>
</tr>
<tr>
<td>7</td>
<td>2</td>
<td>Endo</td>
<td>6, 1, 2, 3, 4, 5</td>
<td>31</td>
<td>Em</td>
<td>UWRS</td>
<td>0</td>
<td>3 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>8</td>
<td>3</td>
<td>Endo</td>
<td>10, 1, 2, 3, 4, 5</td>
<td>34</td>
<td>El</td>
<td>HYS</td>
<td>0</td>
<td>3 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
<tr>
<td>9</td>
<td>2</td>
<td>Endo</td>
<td>8, 1, 2, 3, 4, 5</td>
<td>34</td>
<td>El</td>
<td>UWRS</td>
<td>2</td>
<td>2.5 hrs</td>
<td></td>
<td>Free UB wall</td>
</tr>
<tr>
<td>10</td>
<td>4</td>
<td>Endo</td>
<td>7, 1, 2, 3, 4, 5</td>
<td>34</td>
<td>El</td>
<td>HYS</td>
<td>0</td>
<td>3.5 hrs</td>
<td></td>
<td>Crossing vessels to UB</td>
</tr>
</tbody>
</table>

**TABLE 1: Summary of results**

LSCS: Lower segment Cesarean section

CSP: Cesarean scar Pregnancy

GA: Gestational Age

1. Loss of Hypoechoic line unmeasurable space

2. Vascularity

3. Equal signs—crossing/ bridging vessels.


5. Fazari’s sign

Exo: Exogenous

Endo: Endogenous

HYS: hysterectomy

UWRS: uterine wall reconstruction surgery

UB: urinary bladder

Em: emergency

El: elective

**Discussion**

CSP is considered as a precursor of PAS, increased incidence over the recent years may be secondary to the increased cesarean section rates globally and especially in middle east region, due to cultural background. [3,7,8,9]

Cesarean scar pregnancy itself and morbidity adherent placenta/(MAP)/placenta accreta spectrum (PAS) is often associated with maternal and fetal morbidity and mortality with complications of major obstetric hemorrhage, hysterectomy, surgical complications, preterm birth and fetal death. [1,2,3,5,8,10,11,12]
Concerns of raising incidence of these conditions, globally, especially in middle east region, necessitates to establish clear strategies and protocol for diagnosis, monitoring, management and follow up in tertiary center with expertise and training. [5,6]

It was recommended that in women who choose expectant management, cesarean delivery, should be performed between 34 and 35 weeks of gestation. If cesarean scar pregnancy is complicated with morbidly adherent placenta, most frequent therapeutic approach is removal of uterus. [1] The cesarean section and Hysterectomy should be carried out by an appropriately experienced operator, provided that the patient is hospitalized in a tertiary unit, and a Senior Anesthetist, Urologist and Vascular Surgeon. An intensive care unit and a hospital transfusion laboratory capable of obtaining blood products immediately are required as well. [6]

There is no guidance on best practices for expectant management of CSP, and there is considerable variation, globally. Many studies have reported significant maternal morbidity in cases of CSP, managed expectantly, largely relating to hemorrhage and cesarean hysterectomy from placenta-accreta spectrum. However, high live birth rates are also reported in some studies. [13,14]

There are limited data relating to management and maternal outcomes for expectant management of CSP continuing as PAS/MAP, with the literature being confined to case reports and retrospective cohort studies. [1,12]

Considering our studies, we studied a small group of population in women who refuse termination of pregnancy, due to impact of cultural background in UAE, and patients who are highly motivated to proceed with the pregnancy and expectant management, in spite of knowing all the risks of CSP and PAS. The sonographic signs included in this review and the outcomes described are helpful in counselling women managed expectantly about their prognosis.

An online-based search of PubMed and Cochrane Library Databases were used to gather studies including women diagnosed with a CSP who were managed expectantly and continued as PAS, to compare our outcomes. [1,3]

Our studies, outcomes are in harmony with studies conducted by He F, Li JQ, Tang XL, Lin Y, Su CH, Chen DJ, and Zhonghua Fu Chan Ke Za Zhi, and G Calì, I E Timor-Tritsch.

Both studies are, published in "Pubmed series" in NIH, in 2017 and 2018, supporting the expectant management of CSP continuing as MAP/PAS, when these cases are managed in tertiary center with expertise management, with less mortality and morbidity and also correlating the fact, that CSP is the precursor of PAS/MAP, especially CSP Type1 (endogenous) and type2 (exogenous) but limitations being, these studies were also done in small population. [7,8,9,12]

Another multicenter studies done by Acta Med Okayama, authors published in pubmed series In august 2021, says that the optimal management of CSpS remains unclear; they investigated the clinical courses of CSpS diagnosed and treated at perinatal institutions in the Chugoku and Shikoku regions of Japan. They enrolled 60 women diagnosed with CSP at 21 institutions from January 2006 to December 2015. Of the 60 women diagnosed with CSP, 57 were treated. In Majority pregnancy was terminated (48 women) and continued in 9 women. Thirteen women underwent transabdominal hysterectomy; they experienced no postoperative complications or allogeneic blood transfusion. Among 9 women who chose to continue with their CSP, 7 successfully delivered newborns, 2 had uterine ruptures in the second trimester, and all women required transabdominal hysterectomy, which infers, successful outcome in majority among the women who continued the pregnancies with small risk of uterine rupture, which agrees with our study results. [10]

In contrary, to the recent studies done by Helena C. Bartels and Beatriz Silva, which was published in 2023, European journal of obstetrics and gynecology & reproductive biology their studies says, patients who had expectant management of CSP, outcomes showed (20,1%) had a miscarriage, (8,5%) suffered fetal death. uterine rupture occurred in 9.9% of women in the first or second trimester. For those cases that progressed to the third trimester, 70% developed placenta percreta and 40% experienced severe bleeding. (25,8%) had a term delivery and (41,8%) patients had a preterm birth, out of which (13,9%) delivered before 34 weeks of gestation. In (52,6%) patients, a hysterectomy was performed. [14]

Our studies revealed [ Even though small population study] 90% delivered at 3rd trimester (around 34 weeks' gestation) 10 percent delivered in 2nd trimester at 27 weeks (preterm birth)

70% had cesarean hysterectomy, 30 percent had retained the uterus and had uterine wall reconstruction surgery in cases of focal accrete out of all surgeries, 30 percent were done as emergency and 70 percent as elective. Live birthrate in our study was 100 percent, however 30 percent neonates were admitted to NICU, for prematurity. Only 40% needed blood transfusion. 40 percent were exogenous, 60 percent endogenous types of CSP which continues to progression of MAP/PAS. There were no miscarriages, no maternal deaths, and no significant obstetric complications like rupture of uterus, major obstetric hemorrhage.

The best and first step in dealing with CSP, MAP/PAS mandates setting clear strategies for early diagnosis, referral, patient selection, follow up, care, and management. The surgical management choices may be
considered according to available expertise and geographical and individual circumstances. Preoperative assessment and counseling, intraoperative technique and plan of care, and postoperative follow up and education is the pillars for successful outcome. Furthermore, larger studies, more research are needed for definitive conclusions and to determine the risks of expectant management of CSP and PAS/MAS. [6]

Conclusions
Even though CSP is potentially life threatening condition because of serious complications like placenta accreta if continued [especially in CSP type 2], however expectant management is possible with close monitoring in a tertiary center, under multidisciplinary team with expertise and also strictly adhering to clinical protocols during antenatal period and surgery with accurate surgical techniques to reduce obstetric hemorrhage and by reducing overall mortality and morbidity. Further studies are needed for definitive conclusions and to determine the risks of expectant management.

Additional Information

Disclosures
Human subjects: Consent was obtained or waived by all participants in this study. Animal subjects: All authors have confirmed that this study did not involve animal subjects or tissue. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

References
1. Cesarean scar pregnancies: Experience of 60 cases . J Ultrasound Med. 2015;601-10. 10.7863/ultra.54.4.601
4. 2019:1-45. 10.1111/1471-0528.15586
6. ISSN::1756-222B. 10.3845/GJOFNM.416215
2023. 10.1016/j.progy.2023.102353