CAESAREAN SCAR PREGNANCY: A MATERNAL NEAR- MISS IN THE SANDEMA DISTRICT HOSPITAL, GHANA.

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Abstract -

Introduction: An ectopic pregnancy located in a caesarean scar could have more catastrophic sequelae due to abnormal placentation and early invasion of the myometrium. It has a high possibility of Placenta previa and antenatal uterine rupture as the pregnancy progresses with gestational age. It is very high risk for severe maternal morbidity and associated high maternal mortality.

The case: We presented a case reporting a few weeks after missing her period to the Sandema District Hospital with bleeding per vaginum and a positive urine pregnancy test. The initial diagnosis was a threatened miscarriage with a differential of possible

cervicoisthmic pregnancy after ultrasound scan was done. She was managed conservatively as a case of threatened miscarriage to be reviewed weekly but she defaulted to review and presented four months later with hemodynamic instability after she collapsed at home at 18 weeks gestation. Laparotomy with wedge resection and reconstruction of the previous caesarean section scar was done on account of ruptured uterus and placenta percreta at 18 weeks.

Conclusion: In patients with previous caesarean scar, caesarean ectopic pregnancy should also be suspected when managing them for threatened abortions even in primary care facilities of low income countries.

Keywords: Caesarean scar, ectopic pregnancy, ruptured ectopic, threatened abortion, near-miss

Introduction

Caesarean scar pregnancy is a rare type of ectopic pregnancy occurring in 1/1800 to 1/2500 pregnancies 1,2,3. It results from the implantation and development of the blastocyst in the scar of a previous caesarean section 4,5. It is considered more aggressive than placenta previa or accreta due to the early invasion of the myometrium causing very high risk for uterine rupture with all of its related complications.

It is important because it is on the ascendency as a result of the increasing prevalence of caesarean section scars, improvement in transvaginal sonography and the use of assisted reproduction^{3,4,6}. It is associated with adverse maternal and fetal outcomes if not recognized early such as major hemorrhage, morbidly adherent placenta and placenta praevia, uterine rupture and growth restriction⁶. Early diagnosis leads to prompt management and improves the outcome by allowing preservation of future fertility. Furthermore it has a tendency of being missed especially in developing

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countries with inadequate sonographic equipment and expertise as well as high risk of defaulting due to inadequate health systems⁷. It may then lead to maternal mortality especially where the surgical expertise/ blood transfusion is deficient. The odds of these deficiencies are worse in the rural parts of SubSaharan Africa.

Since its initial description as a uterine scar sacculus⁸, it has generated a lot of discussion with varying opinions as to its exact aetiology and the best mode of treatment. It is thought that Caesarean Scar Pregnancy is due to damage to the endometrium and myometrium by previous cesarean section leading to microscopic tracts through which the trophoblasts burrow into the vesicouterine fold ^{4,9}.

Vial classified caesarean scar pregnancy into two ¹⁰: Type 1 is endophytic, abserosal and grows towards the endometrial/endocervical canal. It is associated with a myometrial thickness of more than 2 mm separating the gestational mass from the bladder. It carries a lower risk of penetrating the serosa. Type 2 is exophytic and grows towards the serosa with a reduced myometrial thickness (<2mm) separating it from the bladder or peritoneal cavity. It has a high risk of penetrating the serosa and bladder manifested by uterine rupture or placental percreta.

The main risk factor is a previous caesarean scar^{10,11}. The number of caesarean sections has not been established to correlate with the risk of caesarean scar pregnancy¹¹. Caesarean section for breech stands out as a significant risk factor for subsequent scar pregnancy¹².

Presentation may range from asymptomatic in the endophytic type to initial painless vaginal bleeding with pain of increasing severity as the blastocyst burrows into the myometrium in the exophytic type. Physical examination may vary dependent on the degree of disruption of the myometrium and hemodynamic status. Peritoneal irritation may result from ruptured uterus or percreta as a result of late stage disease.

As with other types of ectopic pregnancy, slow rise in Beta Human chorionic gonadotrophin may not clearly define this disease¹³. Transvaginal sonography with color flow Doppler gives diagnosis with a sensitivity of 84.6%³ in early disease and Magnetic resonance imaging is the gold standard imaging modality². The criteria for diagnosis is as enumerated by Jayram and colleagues in Table 1 ¹².

Table 1: Ultrasound criteria for diagnosis of caesarean scar pregnancy (CSP)¹²

- 1. Empty uterine cavity and closed and empty cervical canal
- 2. Placenta and/or a gestational sac embedded in the scar of a previous caesarean section
- 3. A triangular/round or oval-shaped gestational sac that fills the niche of the scar
- 4. A thin or absent myometrial layer between the gestational sac and the bladder
- 5. Yolk sac, embryo and cardiac activity may not be present
- 6. Evidence of functional trophoblastic/placental circulation on colour flow Doppler examination, characterized by high velocity and low impedance blood flow

7. Negative 'sliding organs' sign

Diagnostic laparoscopy may only be revealing when the bulge has occurred in the vesicouterine fold, which may not be in early disease. Operative laparoscopy should not be attempted during diagnostic laparoscopy if it had not been suspected earlier and appropriate preparations made given the risk of significant hemorrhage.

Management involves accurate diagnosis coupled with appropriate treatment. Conservative management is largely discouraged as a result of the morbidity and mortality associated with the condition with increasing gestational age³. Medical treatment may be systemic methotrexate or intra-lesion injection of embryocides (KCl or methotrexate)⁴. This may be used alone or as prelude to surgical evacuation when medical methods fail¹⁴. Surgical evacuation may be by transvaginal⁵, hysteroscopic, laparoscopic or open methods with the ancillary application of bilateral hypogastric artery ligation, tourniquet, interventional radiologic methods of uterine artery embolization or balloon placement in order to reduce blood loss.

The risk factors for recurrence include myometrial thickness of less than 5mm, type 2 scar pregnancy, bulging into the uterovesical fold, delivery in a rural

community Hospital, irregular vaginal bleeding and abdominal pain in the previous scar pregnancy¹².

The case

A 25 year old G2P1 reported to the Sandema District Hospital a few weeks after missing her period with bleeding per vaginam and had positive urine pregnancy test. She had a caesarean section 9 months earlier in the same Hospital on account of abruptio placentae, fetal demise and an unfavorable cervix. She was otherwise well. The abdomen was soft and moved with respiration with no tenderness or mass palpable. Vaginal examination revealed moderate vaginal bleeding with a normal looking cervix and closed external os with no cervical motion tenderness. A transabdominal ultrasound showed a small gestational sac in the lower uterine segment (Fig.1).



Fig. 1 transabdominal scan after bleeding per vaginam. UB=urinary Bladder, GS=gestational sac, UT= uterus

A diagnosis of threatened abortion or cervicoisthmic pregnancy was made and patient admitted and managed conservatively. The bleeding and abdominal pain stopped and after 2 days, patient was counselled on the possible outcomes and scheduled for review weekly. She however defaulted review and presented 4 months later after she collapsed at home. Assessment revealed a moderately pale woman with pulse= 120bpm blood pressure=100/70 mmHg and shock index=1.2. Abdomen was tender with rebound tenderness and guarding. Ultrasound showed fluid in abdomen with a live fetus in between the bladder and anterior lower uterine segment with no intervening myometrial tissue. Patient was counselled, resuscitated and laparotomy performed. The family was initially in denial and wished for continuation of the pregnancy at all costs. Persistent counselling while resuscitation continued finally yielded acceptance. The findings at laparotomy were: hemoperitoneum of 2 litres, normal tubes and ovaries, bulky uterus of about 14 weeks sized with a ballooned lower segment and placental tissue and fetal membranes penetrating the serosa on the right aspect of the caesarean scar and bleeding actively (Fig. 2).

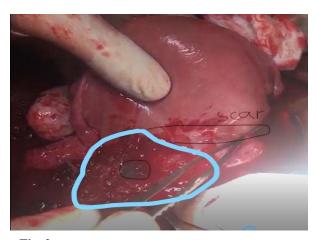


Fig. 2: Photograph of uterus at laparotomy with caesarean scar, vesicouterine bulge and placenta tissue and fetal membranes rupturing through the right aspect of the scar.

The placenta was morbidly adherent and with extensive placental stands throughout the entire scar. The fetus and membranes dissected through the vesicouterine fold and were covered by only that peritoneum beyond the left limit of the rupture. The resection of the gestation was approached by dissecting off the vesicouterine fold followed by encaul delivery of the 18 weeks fetus which died after 10minutes. The placenta was then delivered piecemeal after application of a tourniquet below the gestational mass and surrounding tissues to minimize haemorrhage. Hemostasis was secured completely with ligation of the right uterine artery. The scar was reconstructed using continuous first layer suturing and interrupted second layer suturing (Fig. 3).



Fig. 3: lower segment of uterus after wedge excision and reconstruction of caesarean scar.

She was transfused 3 units of whole blood intraoperative and postoperatively. Post operative condition of patient was satisfactory and patient was discharged on postoperative day 3 when the full blood count report came back normal.

Discussion

The paucity of sonographic skill and equipment has affected the timing and accuracy of diagnosis of caesarean scar pregnancy in developing world⁴. In the initial presentation of the patient, a low implantation was noted on the ultrasound. However, the lack of transvaginal approach as well as doppler limited the value of that finding especially in the environment of vaginal bleeding.

The gestation (round) was in the region of the caesarean scar but could not have been used exclusively for the diagnosis since this could represent a low implantation or a miscarriage in process. A thin myometrial layer was seen between the gestational sac and the bladder. Functional trophoblastic tissue could not be demonstrated in the absence of Colour flow Doppler facilities. The negative sliding organ sign could not be used to distinguish the cervicoisthmic pregnancy from a miscarriage in process due to the absence of transvaginal sonography. With such initial diagnostic challenges, close follow up of the patient with more regular visits for review could have presented an opportunity for early confirmation of the diagnosis but as it turned out this patients failed to follow up for her weekly reviews.

The initial diagnosis in this case was a threatened miscarriage with a differential of cervicoisthmic pregnancy. Patient was admitted and upon cessation of bleeding, she was discharged and scheduled for weekly reviews with repeat ultrasound scans since neither Doppler sonography nor was magnetic resonance imaging was immediately available for further investigation. Unfortunately she defaulted partly because of fears of a termination of her pregnancy given the fact that the previous pregnancy ended in a fetal demise about nine months previously due to placental abruptio. These fears were born out of the effect of a pronatalist culture on the victim of adverse obstetric outcome and it is evidenced by the resistance for the definitive abortive procedure¹⁵. Delayed intervention accounted for the progression of the disease with worsening complication risks. A high index of suspicion especially in populations with the low health seeking behavior in the rural parts for developing countries should increase surveillance by admission until confirmation of complete abortion in situations of threatened abortion with the gestational sac in the cervicoisthmic region. However bed availability may limit the realization of this ideal necessitating the individualization of care in such situations. In a recent report from the Korle Bu Teaching Hospital, a tertiary Hospital, with similar initial management, the patient presented to the facility out of schedule because of better health seeking characteristics and better counselling¹⁶. In both patients, there was the diagnostic difficulty of threatened, cervical or caesarean scar pregnancy. Availability of transvaginal sonography helped in improving precision in the case from the teaching Hospital¹⁶.

The management options that have currently been explored include medical, surgical (transvaginal⁵, laparoscopic and open) even though no consensus has been reached for determining the optimal method for a particular situation¹⁷. Medical management with Methotrexate (local injection and systemic⁴) could have been contemplated at the initial presentation but not in the latter presentation. Systemic methotrexate is frequently associated with high failure rates even though its combination with aspiration and local injection has been commendable in the regression rate of the trophoblastic tissues¹⁸. Laparoscopic and transvaginal excision of the gestation could have been used in the initial presentation if the skill and equipment were available. None of these was done because of the stage at which diagnosis was made.

In our patient with hemodynamic instability, ruptured uterus and placenta percreta, the best approach was the open wedge resection and reconstruction of the scar^{3,17}. This was similar to the treatment modality used in the case from the Korle Bu Teaching Hospital¹⁶ as well as that by Deepika and colleagues¹⁹. It also bore morphological similarity with the initial description by Larsen and coworkers despite a later diagnosis⁸. This modality is favoured by the shorter duration of follow up and lower recurrence rate¹⁹. The availability of the surgical skill needed for definitive care at laparotomy significantly improved the outcome in this case.

Our patient has some risk factors for recurrence (primary surgery in a rural Hospital, type 2 caesarean scar pregnancy at an advanced stage with no myometrium intervening, intermittent pain and bleeding in the index pregnancy ¹²). However the scar reconstruction in our treatment modality is likely to confer a lower recurrence risk¹⁹. We have therefore established the linkage though the district public health team for early detection in subsequent pregnancies for appropriate management.

Conclusion

Caesarean scar pregnancy is a recognized long term complication of caesarean section and its incidence is increasing worldwide with the rural communities of developing world not being **spared.** In patients with previous caesarean scar, caesarean ectopic pregnancy should also be suspected when managing them for threatened abortions even in primary care facilities of low income countries. A high index of suspicion with the requisite imaging, surgical expertise and equipment will go a long way to improve the outcome in the management of this rare type of ectopic pregnancy.

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Conflict of interest

None of the Authors has any conflict of interest to declare

References

- 1. Seow K-M, Huang L-W, Lin Y-H, Lin MY-S, Tsai Y-L, Hwang J-L. Cesarean scar pregnancy: issues in management. Ultrasound Obstet Gynecol. 2004;23:247–253.
- Jurkovic D, Hillaby K, Woelfer B, Lawrence A, Salim R, Elson CJ. First-trimester diagnosis and management of pregnancies implanted into the lower uterine segment Cesarean section scar. Ultrasound Obstet Gynecol. 2003;21:220–227.
- Rotas MA, Haberman S, Levgur M. Cesarean scar ectopic pregnancies: etiology, diagnosis, and management. Obstet Gynecol. 2006;107:1373– 1381.
- Godin PA, Bassil S, Donnez J. An ectopic pregnancy developing in a previous caesarian section scar. Fertil Steril. 1997;67:398–400.
- Mackesy MM, Forris J, Chick B, Chauhan NR, Mandell JC, Khurana B. Visual Diagnosis in Emergency Medicine. J Emerg Med [Internet]. 2014;46:685–686. Available from: http://dx.doi.org/10.1016/j.jemermed.2013.11.089
- Timor-tritsch IE, Monteagudo A. Unforeseen consequences of the increasing rate of cesarean deliveries: early placenta accreta and cesarean scar pregnancy. A review. YMOB [Internet]. 2012;207:14–29. Available from: http://dx.doi.org/10.1016/j.ajog.2012.03.007
- LaGrone L, Sadasivam V, Kushner A, Groen R. A review of training opportunities for ultrasonography in low and middle income countries. *Trop Med Int Health*. 2012;17:808–819.
- 8. Larsen J V, Solomon MH. Pregnancy in a uterine scar sacculus--an unusual cause of postabortal haemorrhage. A case report. *S Afr Med J.* 1978;53:142–143.
- Regnard C, Nosbusch M, Fellemans C, Benali N, Rysselberghe MVAN, Barlow P, Rozenberg S. Cesarean section scar evaluation by saline contrast sonohysterography. Ultrasound Obs Gynecol 200. 2004;23:289–292.
- Vial Y, Petignat P, Hohlfeld P. Pregnancy in a Cesarean scar. Ultrasound Obstet Gynecol [Internet]. 2000;16:592–593. Available from: https://doi.org/10.1046/j.1469-0705.2000.00300-2.x
- 11. Ash A, Smith A, Maxwell D. Caesarean scar pregnancy. BJOG. 2007;114:253–263.
- 12. Jayaram PM, Konje J. Caesarean scar ectopic pregnancy: diagnostic challenges and management options. *Obstet Gynaecol*. 2017;:13–20.
- 13. Shepherd RW, Patton PE, Novy MJ, Burry KA. Serial beta-hCG measurements in the early detection of ectopic pregnancy. *Obstet Gynecol*. 1990;75:417–20.

- 14. Fadhlaoui A, Khrouf M, Kh K, Nouira K, Chaker A, Zhioua F. Successful Conservative Treatment of a Cesarean Scar Pregnancy with Systemically Administered Methotrexate and Subsequent Dilatation and Curettage: A Case Report. 2012;2012.
- 15. Heitlinger A. Pronatalism and women's equality policies. *Eur J Popul*. 1991;7:343–375.
- 16. Asah-opoku K, Oduro NE, Swarray-deen A, Mumuni K, Koranteng IO, Senker RC, Rijken M, Nkyekyer K. Case Report Diagnostic and Management Challenges of Caesarean Scar Ectopic Pregnancy in a Lower Middle Income Country. Case Rep Obstet Gynecol. 2019;2019.
- 17. Lin S, Hsieh C, Tu Y, Li Y, Lee C, Hsu W, Shih J. New ultrasound grading system for cesarean scar pregnancy and its implications for management strategies: An observational cohort study. PLoS One. 2018;9:1–14.
- 18. Pędraszewski P, Właźlak E, Panek W, Surkont G. Cesarean scar pregnancy a new challenge for obstetricians. J Ultrason [Internet]. 2018/03/30. 2018;18:56–62. Available from: https://pubmed.ncbi.nlm.nih.gov/29844942
- 19. Deepika, Taru Gupta, Wahi Sonika. A Rare Case Report of Caesarean Scar Ectopic Pregnancy. *J Clin Diagnostic Res.* 2017;11:10–1.