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Case Report

Successful management of a triplet heterotopic caesarean scar pregnancy in spontaneous conception

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ABSTRACT

A cesarean scar ectopic pregnancy -CSEP is a fairly uncommon presentation wherein the conceptus is implanted at the exact scar site of the previous cesarean section deep in the myometrium. Given the relatively rare incidence of CSEP establishing a diagnosis of CSEP can be challenging current standards of therapy have been derived from data obtained from a limited number of patient's management options for CSEP range from medical line of treatment to surgical interventions such as sonography guided injections to laparoscopic excision or laparotomy or combination of these modalities. Herein we report a rare case of triplet pregnancy with one gestation sac implanted at the site of lower segment scar diagnosed on transvaginal ultrasonography along with MRI who was managed successfully with systemic methotrexate.

Keywords: Methotrexate, MRI, Scar ectopic

INTRODUCTION

Due to the increasing number of Caesarean sections performed escalating prevalence of Caesarean scar ectopic pregnancy -CSEP has been noted in literature. Implantation of the blastocyst and subsequent development of the gestation sac within a previous caesarean section scar is extremely rare, although some reports suggest incidences of 1:1800 to 1:2226 pregnancies.^{1,2} The mechanism behind a scar ectopic is not fully understood but is thought to involve invasion of the blastocyst deep into the myometrium via a microscopic channel between the Caesarean section scar and the endometrial canal.³ The developing pregnancy is then completely surrounded by myometrium and fibrous scar tissue and has no contact with the endometrial cavity. The frequency of spontaneous heterotopic pregnancies is reported as 1:10,000 to 1:50,000, but evidence suggests

that assisted reproductive technologies may increase this risk up to 1%.⁴ Caesarean scar pregnancy coexisting with intrauterine gestation is extremely rare. Multiple pregnancies are associated with hyperplacentosis which further increases risk of abnormal placentation. Recognition of scar ectopic on ultrasound is essential, since any delay in diagnosis may result in uterine rupture, haemorrhage and subsequent hysterectomy with loss of future fertility. Evidence also suggests that scar ectopic pregnancies, if untreated, may evolve into morbidly adherent placenta.⁵

CASE REPORT

A 32-year-old Gravida 3 Para1 was admitted with complaints of 2 months of amenorrhoea with pain in abdomen and bleeding per vaginam. Her 1st pregnancy was a caesarean section 6 years ago in view of fetal distress and 2nd pregnancy was terminated in 18th week of

geatation for anencephaly with history of curettage for retained products of conception. Her medical history was unremarkable. Physical examination showed no abdominal tenderness. Speculum examination revealed a normal cervix with minimal bleeding and without any cervical dilatation her vital parametres were stable and on pervaginal examination uterus was about 8 weeks in size with internal os closed and fornices clear. On investigation her hemoglobin was found to be 13.2 g/dL and serum beta hCG was 4408.8mIU/mL. A TVUS was advised which showed two gestational sacs one of which was a 6.2weeks (1.46cm) Myometrial pregnancy with fetal bradycardia. The other gestational sac was 5.2weeks (7mm) without a fetal pole. There was no free fluid in Douglas pouch and no adnexal pathology was observed as shown in Figure 1 In view of overlying myometrial thickness of only 1.8mm MRI was done to confirm the diagnosis of scar ectopic pregnancy.

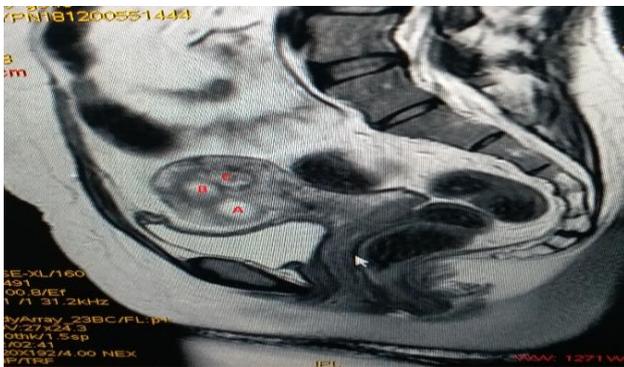


Figure 1: Trichorionic pregnancy on MRI.



Figure 2: Thinned out myometrium on MRI.

Magnetic resonance imaging showed a trichorionic pregnancy as shown in Figure 2 with the largest gestational sac (Sac A) being intra-myometrial at the site of previous caesarean section scar measuring 3.8 x 2.1 x 1.9cms with no fetal pole. The other two empty gestational sacs located cranially (Sac B) measuring 1.9 x 1.4 x 1.3cm and (Sac C) 1.7 x 1.4 x 1.4cm.

The myometrium in the anterior uterine wall corresponding to the lower uterine segment was thinned out and measured 3mm in maximum thickness as shown

in as shown in Figure 2 marked with an arrow. Based on these findings a definitive diagnosis of CSEP was made.



Figure 3: Two gestational sacs disappeared and scar ectopic size reduced.

The patient was informed of all possible treatment options and complications and adequately counselled. Her baseline blood investigations like CBC, RFT, LFT were normal. Because of the aforementioned findings, medical management was opted for this patient. A single dose of intramuscular methotrexate was given at a dose of 1mg/kg followed by an intramuscular dose of Leucovorin at a dose of 0.1mg/kg on the following day. Serial ultrasonography findings revealed a shrinking of all gestational sacs with a falling level of B-hCG. The patient's B-hCG level dropped to 17.2 four weeks after the medication. After 8 weeks of systemic methotrexate injection TVUS showed the scar ectopic measuring 1.1x0.8cm and the other two gestational sacs had disappeared as shown in Figure 3. The patient's B-hCG level dropped to 17.2 four weeks after the medication.

DISCUSSION

The first case of CSEP was reported in 1978.⁶ Literature search has shown barely 19 cases of CSEP between 2001 and by 2007, Only 1000 cases have been reported as yet.⁷ It is probable that implantation at the scar site occurs due to defects in the scar in the form of microtubular tract which develops due to poor healing of the previous trauma caused by previous D and C, LSCS, myomectomy, and manual removal of placenta.⁸ Women who elect for elective caesarean section for abnormal presentation are at increased risk for CSEP possible explanation is poor formation of lower uterine segment requiring relative high incision on lower uterine segment.⁹ Different types of scar ectopic pregnancies are noted. Type I is caused by implantation in the prior scar with progression towards the cervico-isthmic (in prior caesarean section) space or the uterine cavity as was seen in our patient. Type II is due to implantation deep into scar tissue infiltrating full thickness of the myometrium which is most dangerous.¹⁰ Diagnosis depends on symptoms, clinical manifestation, history of previous scar, serum β hCG level, trasvaginal sonography (TVS).

Women with CSEP may present with slight vaginal bleeding, sometimes it may be an incidental finding in asymptomatic women. Haemodynamic instability with acute abdominal pain in a suspected case of CSEP strongly suspects rupture with intrabdominal bleeding.¹¹ Transvaginal sonography is often used for diagnosis. Criteria for the diagnosis of CSEP proposed by Timor-Tritsch are Normal endometrial thickness, gestational sac situated in the anterior wall of uterus corresponding to the scar site of the previous caesarean. Doppler ultrasound demonstrating functional trophoblastic tissue at the site of implantation at the scar. In less than 8 weeks gestation, a triangular shaped gestational sac filling the scar. Cervical canal that is closed and empty. Observation of fetal pole and/or yolk sac with or without heart activity and absence or deficiency of a healthy myometrium between the bladder and the gestational sac.¹²

Magnetic resonance imaging (MRI) is also a useful adjunct for the diagnosis of CSEP. It is especially useful in cases where accurate diagnosis of CSEP by USG is difficult. Differential diagnosis-inevitable abortion with low lying gestational sac and cervical ectopic pregnancy may be considered.¹² In an inevitable abortion the gestational sac is often irregular and located within the uterine cavity and with absent or minimal colour Doppler flows. When gentle pressure is given at the level of the internal cervical os by ultrasound probe may displace the gestational sac. This is known as 'the sliding sign'.¹³ It is absent in CSEP and cervical ectopic pregnancy. In cervical ectopic pregnancy, gestational sac is present near to cervix or inside the cervix with ballooning of cervical canal. On USG doppler flow is good and sliding sign is negative. There is no consensus on treatment of scar ectopic pregnancies and it depends, to a degree, on the gestational age. Furthermore, the minute channel in which the scar ectopic embedded in the first place will still be present and may lead to recurrent scar ectopic pregnancy.¹⁴ Management depends on initial symptoms, fertility preservation, compliance for prolonged follow up, surgical risk factors, gestational age, serum beta HCG levels, Size of CSEP, myometrial thickness and viability. Presence of interventional radiology facility and surgical expertise also influence management options. In asymptomatic woman with nonviable CSEP with decreasing HCG levels, expectant management has been reported successfully.¹⁴ Decision should be carefully balanced with risk systemic methotrexate may be successful for treating early gestations although regression can take a long time and there is still the risk of uterine rupture, need of laparotomy and hysterectomy any time during pregnancy should be clearly discussed and documented methotrexate is drug of choice for medical management. Woman should be counseled for prolonged follow up, repeated beta hCG testing, possibility of surgical intervention including hysterectomy.¹⁵ As was done in our patient Systemic methotrexate regimen is more likely to succeed when initial serum beta hCG level is less than 5000 IU/L and gestational age less than 8 weeks. Local injections of

methotrexate, KCL, hyperosmolar glucose under ultrasound guidance have been used to treat CSEP. combined local and systemic administration of methotrexate is also an alternative option.¹⁶ Bilateral uterine artery chemoembolisation (BUACE) with gel foam in conjugation with systemic or local administration of methotrexate has been used during or after surgical removal of CSEP to prevent bleeding.¹⁷ BUACE is associated with complications as fever, abdominal pain, pelvic infection, amenorrhea from endometrial atrophy and premature ovarian failure. The conventional laparotomy with resection of the ectopic sac along with the previous scar tissue has been replaced, in skilled hands, by a laparoscopic excision alone for complete treatment of CSEP. Patients with an exogenously located CSEP are ideal candidates for laparoscopic intervention. Laparoscopic resection and closure of defect is feasible in exogenous (Type II) CSEP.¹⁸ Hererotropic pregnancies with one sac in the previous caesarean scar and other intrauterine sac have been reported.^{18,19} These cases are challenging for diagnosis as well as for management. Its occurrence in conjunction with a viable intrauterine pregnancy and patient's desire to maintain the intrauterine pregnancy makes the therapeutic management more difficult. In our case, based on findings from serial TVUS performed along with serial B-hCG showing a falling trend, keeping the risks of operative procedures in mind, medical management was opted for this patient with a successful outcome.

CONCLUSION

Attention should be given on primary prevention focusing on reduction in the primary caesarean rate. Risk of future CSEP and adherent placenta should be informed while counselling women requesting for caesarean for non medical reasons. Emphasis should be given on good surgical practice like minimum tissue handling good haemostasis during primary caesarean section. Further studies are needed for single layer versus double layer closure of the uterus and incidence of CSEP. An early and timely diagnosis of CSEP increases success rate and decreases complications to a considerable extent.

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