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Case Report

Isthmocele: emerging emergency

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ABSTRACT

An isthmocele, a caesarean scar defect or uterine niche, is any indentation representing myometrial discontinuity or a triangular anechoic defect in the anterior uterine wall, with the base communicating to the uterine cavity, at the site of a previous caesarean section scar. With rise of caesarean sections, incidence of isthmocele and scar ectopics is on rise, being 19 to 88% in caesarean delivery patients. As there are no definitive criteria for diagnosing an isthmocele, several imaging methods can be used to assess the integrity of the uterine wall and thus diagnose an isthmocele. However, transvaginal ultrasound and saline infusion sonohysterography emerge as specific, sensitive and cost-effective methods to diagnose isthmocele. When cervix is encroached as well by ectopic placentation, situation becomes much more alarming and grave. Timely diagnosis and active management of the entity is a must to save life of patient. Area of isthmocele bearing ectopic gestation sac has to be excised whatever be the method, by endoscopy or conventional open method. Previous gestation scar dehiscence or rupture of uterus repair leads to more weakness and impaired integrity of uterine wall. If patient refuses to undergo simultaneous sterilization, recurrence of isthmocele formation and ultimate scar ectopic will be even higher. Here we presented a case of ectopic gestation in isthmocele complicated by its extension to cervix with an associated posterior wall cervical fibroid at same level.

Keywords: Caesarean section, Isthmocele, Ectopic pregnancy, Gestation, Cervical pregnancy, Shock

INTRODUCTION

Isthmocele is a caesarean scar defect (Figure 1). It can be classified as a small or large defect, depending upon the wall thickness of myometrial deficiency. Although usually asymptomatic, its primary symptom is abnormal or post-menstrual bleeding, and chronic pelvic pain may also occur. Infertility, placenta accrete or praevia, scar dehiscence, uterine rupture, and caesarean scar ectopic may also appear as complications of this condition.¹ Concomitant presence of a cervical fibroid nearby further increases vascularity and distorts anatomy thereby raising risk of precipitation of complications, more so of haemorrhages. Patient had twice caesarean section earlier including rupture uterus in last gestation. It was associated with posterior cervical fibroid at same level. This time scar

ectopic gestation extended to upper cervix as well making situation alarming.²

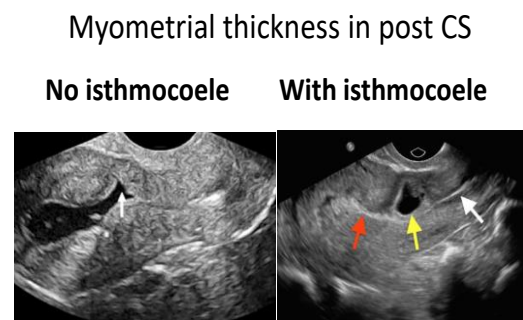


Figure 1: Post CS uterine wall healing patterns.

CASE REPORT

Mrs X, a 33 years, P2+0, both LSCS, had first CS for transverse lie 5 years back, and second CS for breech having features of scar dehiscence and severe anaemia one and half year back. She was then resuscitated with 4 bottles of blood transfusion. Tubectomy was refused by patient and party. She was blessed with 2.8 kg live female child again.

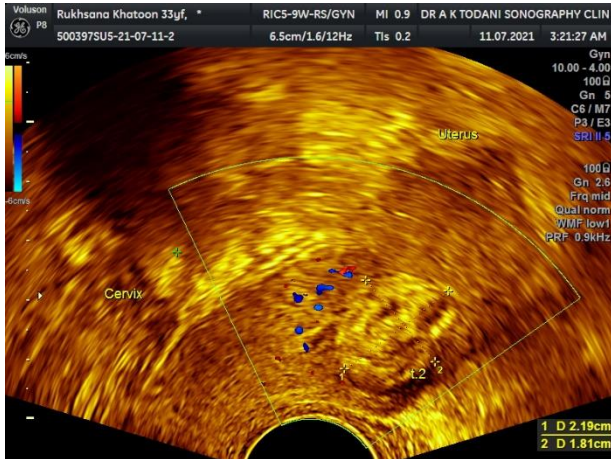


Figure 2: Isthmocele in anterior wall with 2 mm cervical wall thickness and having ectopic pregnancy with cardiac activity.

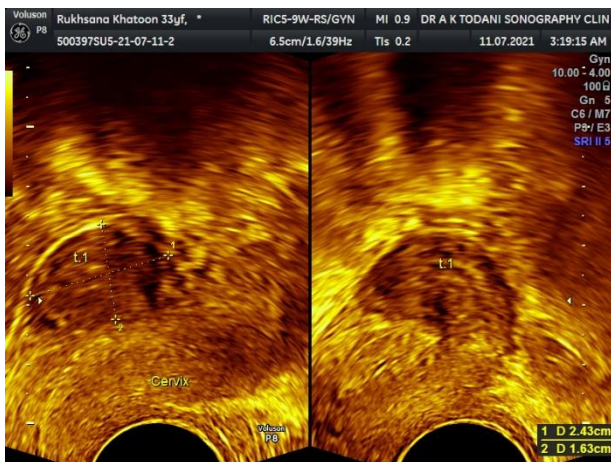


Figure 3: Presence of cervical fibroid in upper posterior wall.

This time she presented with just two weeks overdue periods but profuse vaginal bleeding and shock. Transvaginal sonography revealed presence of isthmocele with presence of gestational sac with cardiac activity encroaching on and embedded in anterior cervical wall as well (Figure 2). One intramural fibroid measuring 2.43×1.63 cm was present in upper cervical wall posteriorly (Figure 3 and 4). Diagnosis of scar ectopic pregnancy presenting as cervical pregnancy as well was made. There was no time to wait. Severe anaemia and

haemorrhagic shock forced us for emergency management for removal of ectopic gestation.³

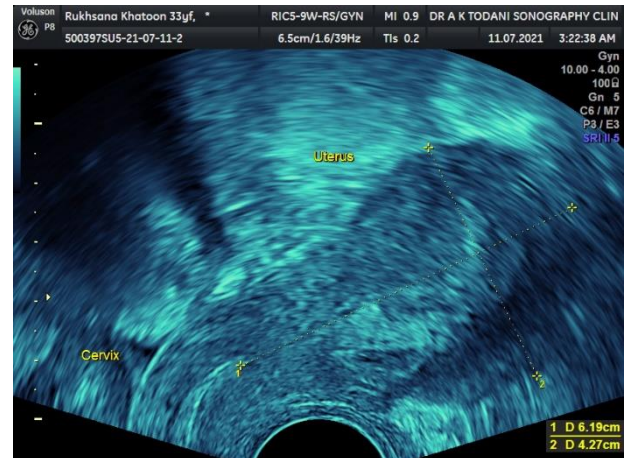


Figure 4: Longitudinal TVS scan of uterus depicting ectopic gestation in isthmocele.



Figure 5: Ectopic gestation sac with embryo inside.

Trial curettage failed to arrest bleeding. Laparotomy was done in a hurry explaining grave prognosis and life risk to patient party.

Abdomen was opened by Phannenstiel incision. Dense abdomino-pelvic and vesico-cervico-peritoneal adhesions were dissected through. Space was created in right adnexal area by dividing right round ligament. Then by utilising Seth's lateral window between broad ligament and uterocervical margin, urinary bladder could be pushed away. At this point, apex of isthmocele got opened up

spontaneously with gush of blood (Figure 5). Gestation sac adherent to cervical wall was present. There was no other way out than doing total hysterectomy with bilateral salpingectomy (Figure 6). Ovaries were healthy and so preserved. Abdomen closed in layers. Five bottles of blood transfusion were given. Patient had uneventful post-operative recovery.



Figure 6: Hysterectomy specimen with anterior isthmocele niche and posterior wall cervical fibroid.

With modernization and increased awareness, more obstetric deliveries are accomplished by caesarean section than normal vaginal route. So, risk of isthmocele formation is on rampant rise. In this case, patient survived after active heroic management of complex entity of live scar ectopic complicated with cervical encroachment of gestation sac leading into intractable vaginal haemorrhage and shock. Here in this patient, urinary bladder formed a ‘flap cover’ over isthmocele bearing ectopic pregnancy.

DISCUSSION

An isthmocele, is a cesarean scar defect, usually lined with endometrium, at the site of a thinned out wall at site of previous cesarean section scar. It can be classified as a small or large defect, depending on the wall thickness of the myometrial deficiency. Although usually asymptomatic, its primary symptom is abnormal or postmenstrual bleeding, and chronic pelvic pain may also occur. Occasionally endometriosis is found to affect this site of isthmocele. Infertility, placenta accrete or praevia, scar dehiscence, uterine rupture, and cesarean scar ectopic pregnancy may also appear as complications of this

condition. The risk factors of isthmocele proven to date include retroflexed uterus and multiple cesarean sections. Nevertheless, factors such as a lower position of cesarean section, incomplete closure of the hysterotomy, early adhesions of the uterine wall and a genetic predisposition may also contribute to the development of a niche. Contributing likely factors are the type of suture used to repair the uterine incision, and whether the uterus was repaired in single or two layers. Exaggerated muscle tissue fibrosis is seen with long absorbing suture materials inducing more foreign body reactions. Evidence suggests double-layer (vs. single-layer) uterine closure decreases the formation of cesarean scar isthmocele and increases residual myometrial thickness (RMT). Additionally, uterine closure with a locked suturing technique may result in greater isthmocele depth and size, and a thinner RMT at the cesarean scar site. To our knowledge however, no study had determined the impact of a locked vs. unlocked double-layer closure technique on isthmocele prevention. Locked sutures, believed to be more hemostatic, may cause enhanced tissue necrosis and impaired wound healing. Interestingly, evidence did not support reductions in operative blood loss with locked closure techniques. The hypothesis was that unlocked double-layer uterine closure can reduce isthmocele formation and thus improve long-term maternal outcomes without impacting operative blood loss or other short-term surgical outcomes. Coexistence of fibroid at same level will widen isthmocele and promote ectopic implantation.

As there were no definitive criteria for diagnosing an isthmocele, several imaging methods can be used to assess the integrity of the uterine wall and thus diagnose an isthmocele. However, transvaginal ultrasound and saline infusion sonohysterography emerge as specific, sensitive and cost-effective methods to diagnose isthmocele. It was often a neglected cause of secondary infertility and implantation failure due to local stagnation of menstrual blood. The treatment included clinical or surgical management, depending on the size of the defect, the presence of symptoms, the presence of secondary infertility and plans of childbearing. In cold cases, surgical management includes minimally invasive approaches with sparing techniques such as hysteroscopic, laparoscopic or transvaginal procedures according to the defect size.⁴ Hysteroscopic resection is preferred for small niche with residual muscle thickness more than 3 mm, niche located lower down can be treated transvaginally, and trans-abdominal approach is preferred for large defects in women desiring future pregnancy.⁵

CONCLUSION

Isthmocele is receiving attention worldwide for last 4 years or more because of it's direct relation with increasing rate of caesarean section. It annoys a clinician when it presents as acute clinical emergency. With availability of ultrasonography in every nook and corner of inhabitation, oligohydramnios, nuchal cord, foetal and or maternal distress are diagnosed more frequently. Result is more

caesarean section and more isthmocele formation. Diagnosis of isthmocele is a diagnostic dilemma; but presentation of clinical emergency like haemorrhage, pain and shock are cumbersome more so when pregnancy encroaches upon cervical wall. Only way out is reduction of caesarean incidence, improving uterine wall closure technique, asepsis promoting epithelialisation of uterine niche and prompt treatment of asymptomatic or symptomatic isthmocele. Probably we have to be more isthmocele minded just like we are ectopic minded.

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