Title: Rupture of Unscarred uterus in a nulliparous in unestablished labour

Nadia Rahman (Obs & Gynae Registrar), Sangeeta Pathak (Consultant Obs & gyane)

Introduction:

Uterine rupture is an extreme life threatening obstetric emergency associated with maternal and fetal morbidity and mortality. Uterine rupture in the absence of previous scar has an estimated incidence of 1 in 5700 to 1 in 20,000.[1]

The identifiable causes of ruptured uterus include previous surgery, physical trauma, multiparty, prolonged labour, augmentation or induction of labour, collagen disorders of collagen and structural uterine abnormalities.

The woman we report was a low risk with uterine rupture in an unscarred and non-labouring uterus at term which is extremely rare. Although there was good maternal and fetal outcome in this case as she was managed in timely manner after she was brought to hospital, this could have been a different outcome if she had not arrived on time. The management of her future pregnancy remains a sizable challenge for clinicians.

Case History:

We report the case of a 23 years old Caucasian nulliparous with an uneventful antenatal course. Her past history comprised of her knees locking and clicking during movement without joints swelling or dislocations at the age of 12. This was investigated without definitive diagnosis.

She presented to us via ambulance reporting lower abdominal pain for 2 hours prior to admission. The pain was sudden onset while trying to open her bowels and was continuously getting worse. On clinical examination her blood pressure was 120/78mmHg with a pulse of 126 beats/minute(bpm), respiratory rate of 18 and oxygen saturation of 100%. Her haemoglobin was 111gms/L, WCC 27.0 x 10 9/L and platelets 199 x 109/L. On abdominal palpation uterus was soft, tender but relaxed in between tightenings with 3/5th palpable cephalic presentation. On auscultation with fetal heart was 90 bpm and Cardiotocography(CTG) was commenced. Vaginal examination revealed that cervical os was 1 cm dilated, with presenting part at station -3. Membranes were artificially ruptured (ARM) and slight blood stained liquor was noticed. A category 1 emergency caesarean section (EMCS) was performed due to fetal bradycardia with the working diagnosis of placental abruption. Moderate amount of hemoperitoneum was noted intraoperatively. A live baby weighing 3160gms was delivered with apgar scores of 2, 6 and 6 at 1, 5 and 10 minutes respectively. The cord gases were arterial pH 7.00 with base excess (BE) of -15 and venous pH 6.97 with BE -15. Placenta appeared normal and complete without evidence of abruption. On exploration, a left sided vertical laceration was noted on the posterior aspect of the uterus, involving the entire thickness arising from left tubal insertion till the proximal part of vagina. This rupture was located 3-4cms medial to the left ovarian vessels. A diagnosis of a spontaneous rupture of uterus in an unscarred uterus was made. Both the EMCS incision and uterine wall rupture were individually repaired with vicryl, flowseal applied to the area of rupture and drain was left in-situ. Her total blood loss during the EMCS was 1700mls. The patient was discharged 4 days later and was seen at 6 weeks follow up to discuss about risks in future pregnancy, timing and mode of delivery.

In view of the rupture of unscarred uterus without labour and history of easy dislocation of her knees, the possibility of Ehlers-Danlos-Syndrome or other connective tissue disorder was considered and she was referred to rheumatologist for further investigations postnatally. After clinical assessment of joint mobility by Brighton scoring (a score of four or more suggests likelihood of joint hypermobility)[2] she was further referred to the National Diagnostic Centre for Ehlers-Danlos. She was tested for Vascular Ehlers-Danlos-Syndrome (COL3A1 gene mutation) and no abnormalities were found.

Discussion:

Uterine rupture is defined as a full thickness tear through the myometrium and serosa with or without expulsion of the fetus from the uterine cavity.

The overall incidence of uterine rupture is 0.05 to 0.086% of all pregnancies.[3] Rupture of a scarred uterus is more common with the overall risk of 0.9% to 1% in a woman attempting vaginal birth after caesarean section(VABC).[4] Incidence of rupture in a previous classical section increases dramatically to 3-6% and 12% if VBAC is attempted.[5] Taylor et al showed in a multicentre study that the risk of rupture in previous LSCS attempting VBAC was higher when induced with vaginal prostaglandins (10.3% vs. 1.1%.[6] The incidence of uterine rupture in women with unscarred uterus undergoing augmentation is extremely low. Cahill analysed a consecutive series of 30874 term primiparous deliveries over a period of 13 years, of which 45% received oxytocin for augmentation without a single case of uterine rupture thus reassuring about the safety of use of oxytocin in primips.[7] A large cross-sectional study indicated that the prevalence in women with unscarred uterus is less than 1 in 10,000.[8] Most of these studies include women who have one of the risk factors; either in labour, on oxytocin or being induced using prostaglandins for obstetric indications.

This serious obstetric complication is particularly higher in developing countries as compared to developed countries.[9] Gaym and Udoma et al reported large numbers of uterine rupture cases as a result of obstructed labour over a 9 year period in Ethiopia(25%) and Nigeria(19%).[10,11]

Over the past few years, the number of both rupture of scarred and unscarred uterus has been observed to be increasing.[12] The presence of contributory factors in a woman with unscarred uterus such as multiple gestation, uterine congenital abnormality, abnormal placentation, drug use, prolonged labour, and even judicious use of oxytocin in labour, mid cavity forceps, internal podalic version may compound the risk of this potentially life threatening obstetric complication.

Due to the urgency of situation and the possibility of fetal loss, time consuming diagnostic tools and imaging facilities have extremely limited use. Assessment of clinical signs remains the gold standard for diagnosis and guides management. However even with this limitation ultrasound, CT and MRI have been used to assess high risk cases.[13]

Treatment options consist of surgical repair or hysterectomy. Surgical repair should be attempted if technically feasible where it can achieve rapid hemodynamic stability and also if there is a desire for future fertility. However the risk of future rupture is significantly higher; 6% with repeat lower segment rupture and 32% with previous upper segment rupture.[14] If uterine repair is not suitable, total or sub-total hysterectomy is the next option, depending on the extension of the tear. There is robust evidence to suggest that sub-total hysterectomy is associated with less operating time, shorter hospital stay and lower morbidity and mortality as compared to surgical repair in selected cases.[15]

Ehler Danlos Syndrome is a heterogenous collection of rare disorders of the connective tissue. The prevalence has been recently estimated to be 1 in 5000.[16] This rarity makes it difficult to estimate the true incidence of complications which include pelvic instability, complicated perineal wounds, rupture of vessels/bowels/uterus and floppy infant syndrome. The more severe complications have been reported in Type IV syndrome. On the whole pregnancy is generally well-tolerated in Type I-III with favourable maternal and fetal outcomes. Studies from Dutch Ehler Danlos Association and American Ehlers–Danlos National Foundation showed no cases of uterine rupture or any other complications.[17,18] In our case there was a high degree of suspicion of connective tissue disorder however this was ruled out after series of investigations.

A high index of suspicion is needed to make the diagnosis of ruptured uterus. A preoperative provisional diagnosis is not critical since delivery is often indicated because of abnormal fetal monitoring patterns, pain or hemodynamic instability. However symptoms may be subtle in some cases. The most common clinical sign, sudden fetal decompensation is reported in 80% cases with bradycardia.[19]The other

symptoms are hyper-stimulation (40% cases), vaginal bleed and abdominal pain. Immediate maternal collapse is rare unless the uterine tear extends into the broad ligament vessels.

In our case we discussed the challenges around her next pregnancy. Inpatient management from 34 weeks versus outpatient management was discussed. Risks of prolonged admission such as hospital acquired infection, thromboembolism and risks to baby e.g. fetal death in the event of rupture (given the additional caesarean section scar), iatrogenic prematurity; respiratory distress syndrome and prolonged SCBU stay were carefully considered. On balance it was thought the best care for future pregnancy would comprise of admission in hospital at 34 weeks, administration of steroids and elective caesarean section by 36 weeks. We agree that offering counselling and a multi-disciplinary team approach in accordance with the local trust policy is the key in managing these complex patients.

Conflict of Interest: None

Declaration of author: Patients written consent was obtained.

References:

1. Zwart JJ, Richters JM, Ory F, et al. Uterine rupture in The Netherlands: a nationwide population-based cohort study. BJOG 2009; 116:1069
2. Brighton PH, Solomon L, Soskolne CL. Articular mobility in an African population. Ann Rheum Dis. 1973; 32: 413-17

3. Eden RD, Parker RT, Gall SA: Rupture of the pregnant uterus: A 53-year review. Obstet Gynecol. 68:671-674 1986

4. Kaczmarczyk M, Sparén P, Terry P, Cnattingius S: Risk factors for uterine rupture and neonatal consequences of uterine rupture: A population-based study of successive pregnancies in Sweden. BJOG. 114:1208-1214 2007

5. Endres LK, Barnhart K: Spontaneous second trimester uterine rupture after classical caesarean. Obstet Gynecol. 96:806-808 2000

6. Taylor DR, Doughty AS, Kaufman H, et al.: Uterine rupture with the use of PGE2 vaginal inserts for labor induction in women with previous cesarean sections. J Reprod Med. 47:549-554 2002

7. Cahill DJ, Boylan PC, O'Herlihy C: Does oxytocin augmentation increase perinatal risk in primigravid labour? Am J Obstet Gynecol 1992; 166: 847-850

8. Miller DA, Goodwin TM, Gherman RB, Paul RH. Intrapartum rupture of the unscarred uterus. Obstet Gynecol 1997;89(5 Pt 1):671-673

9. Nagarkatti RS, Ambiye VR, Vaidya PR: Rupture uterus: Changing trends in etiology and management. J Postgrad Med. 37:136-139 1991

10. Gaym A. Obstructed labour at a district hospital. Ethiop Med J 2002;40(1):11-18

11. Udoma EJ, Asuquot EE, Ekott MI. Maternal Mortality from Obstructed labour in south-east Nigeria: the role of spiritual . Int J Gynaecol Obstet 1999;67(2):103-105

12. Al-Zirqi I, Stray-Pedersen B, Forsén L, et al. Uterine rupture: trends over 40 years. BJOG 2015

13. Hruska KM1, Coughlin BF, Coggins AA, Wiczyk HP: MRI diagnosis of spontaneous uterine rupture of an unscarred uterus. Emerg Radiol. 2006 May;12(4):186-8

14. Ritchie EH: Pregnancy after rupture of the pregnant uterus. A report of 36 pregnancies and a study of cases reported since 1932. J Obstet Gynaecol Br Commonw. 78:642-648 1971

15. Thakur A, Heer MS, Thakur V, et al.: Subtotal hysterectomy for uterine rupture. Int J Gynecol Obstet. 74:29-33 2001

16. Steinmann B, Royce PM, Superti-Furga A. The Ehlers–Danlos syndrome. In: RoycePM, SteinmannB (eds): Connective Tissue and its Heritable Disorders. Molecular, Genetic and Medical Aspects. 2nd Ed. New York: John Wiley, 1993: 351–408

17. Jan Lind, Henk C. S. Wallenburg: Pregnancy and the Ehlers–Danlos syndrome: a retrospective study in a Dutch population Acta Obstetricia et Gynecologica Scandinavica April 2001,Vol 81, Issue 4: 293–300

18. Sorokin Y, Johnson MP, Rogowski N, Richardson DA, Evans MI. Obstetric and gynecologic dysfunction in the Ehlers–Danlos syndrome. J Reprod Med 1994; 39: 281–4

19. Guise JM, Hashima J, Osterweil P: Evidence-based vaginal birth after caesarean section. Best Pract Res Clin Obstet Gynaecol. 19:117-130 2005