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CASE STUDY

SPONTANEOUS MIDTRIMESTER UTERINE RUPTURE DUE TO AN ANGULAR PREGNANCY

*Shaveta Jagdevan, Sachin Mehta, Laljee K. Makwana and Pagi, S. L.

Department of Obstetrics & Gynaecology, Baroda Medical College and SSG Hospital, Vadodara, India

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ABSTRACT

Uterine rupture is a catastrophic complication of pregnancy occurring most commonly during third trimester. Though, uterine rupture in first and second trimesters is rare, spontaneous mid-trimester rupture is even rarer with only four such cases described in English literature. We hereby report an extremely rare case of spontaneous midtrimester uterine rupture due to angular pregnancy. With this case report we intend to emphasize that though angular pregnancy is a rare clinical entity, it should be considered in differential diagnosis of patients presenting with haemoperitoneum in early/mid pregnancy. A knowledge of ultrasonographic features of angular pregnancy can potentially prevent occurrence of a catastrophic consequence.

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INTRODUCTION

Uterine rupture is a catastrophic complication of pregnancy occurring most commonly during third trimester. Uterine rupture in first and second trimesters is a rare occurrence and has been attributed to placenta percreta, (Fleisch *et al.*, 2008) previous caesareans, (Fleisch *et al.*, 2008) previous myomectomy (Barron *et al.*, 1966) and angular pregnancy (Torbé *et al.*, 2012; Kerr and Anderson, 1934; Rigby, 1951; Baldawa and Chaudhari, 2008). Spontaneous midtrimester rupture is extremely rare. We hereby report an extremely rare case where a pregnant patient in second trimester presented with spontaneous uterine rupture and hemoperitoneum due to angular pregnancy. To the best of our knowledge, only four such cases of spontaneously ruptured angular pregnancy in second trimester have been reported in available English literature.

Case Report

A 28 year old gravida 5 para 2 patient presented to our emergency department with acute onset abdominal pain at 14 weeks of gestation. On examination, patient was pale, in hypovolemic shock (cold clammy peripheries, pulse rate-

*Corresponding author: Shaveta Jagdevan,

Department of Obstetrics & Gynaecology, Baroda Medical College and SSG Hospital, Vadodara, India.

128/min, B.P- 80 mmhg systolic) and had abdominal distension and tenderness in bilateral lower quadrants. Pelvic examination revealed closed cervical os with no vaginal bleeding, Uterus was tender on bimanual examination and there was no adnexal pathology. After resuscitation, ultrasonography of abdomen and pelvis was done which revealed gross haemoperitoneum and an intrauterine pregnancy of 15weeks maturity with absent cardiac activity. The source of haemoperitoneum could not be ascertained. On exploratory laparotomy, ~1500 ml blood was present in peritoneal cavity. An enlarged uterus (Fig-1a&b) with distended right cornual region (*in Fig-1a&b) was present. A rent of about 1x1cm with protruding placental tissue was seen on the distended right cornua (arrow head in Fig-1a&b).

Very small size of the rent, extreme friability of its margins and active bleeding from rent site precluded delivery of fetus through the rent. The fetus was thus deliverd by hysterotomy and the rent was repaired. Complete hemostasis was ensured. Thorough peritoneal lavage was done. Patient made uneventful recovery and was discharged on 10th postoperative day. Tubal ligation was not done, as patient did not give consent for the same preoperatively, however, patient was counselled and underwent laparoscopic tubal ligation after two months.

Table 1. The cases of spontaneous mid-trimester rupture of angular pregnancy in literature

TM: Trimester

Authors	Year	Gravidity	Gestational age (weeks)	Site of Rupture	Management
Rigby et al. (1951)	1951	4	2 nd TM	Left cornua	Hysterectomy
Barron et al. (1966)	1966	3	14	Left cornua	Total abdominal hysterectomy+left oophorectomy
Stiller et al. (1991)	1991	3	21	Right fundal region	Hysterotomy
Baldawa et al. (2008)	2008	3	14	Right lateral wall	Subtotalhysterectomy + right salpingectomy
Present case	2012	5	14	Right cornua	Hysterotomy

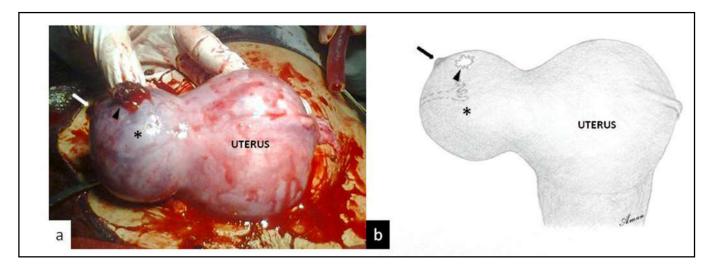


Figure 1. The intra-operative photograph (a) and diagrammatic representation (b) of ruptured angular pregnancy. (*) in figures-a & b indicates distended right cornua and arrow head points at the site of rupture. Arrows in figures- a & b identify the round ligament that has been displaced laterally by the angular pregnancy

DISCUSSION

The term angular pregnancy was first described by Kelly H. in 1898 (Kelly, 1898) as a pregnancy where an embryo implants in lateral angle of uterine cavity medial to the internal ostium of fallopian tube. The term has been at times used interchangeably with interstitial pregnancy however, the two are separate clinical entities (Kerr and Anderson, 1934). Jansen and Elliot, (1981) reviewed 39 cases of angular pregnancy and reported uterine anomalies (septate or bicornuate uterus) as a predisposing factor for angular pregnancy. No other specific risk factors have been mentioned in the literature. Kerr & Anderson, (1934) found three features in all the cases of angular pregnancy viz. pain, lateral distension of uterus in the region of cornua and a tendency to abort, and also described the characteristic uterine distension in angular pregnancy as an elongated distended lateral sacculation at one cornua. Abdominal pain and characteristic uterine distension (*in Figure-1a & b) were present in our patient as well. The fate of angular pregnancy can be uterine rupture, spontaneous abortion or spontaneous correction to normal intrauterine pregnancy. Angular pregnancies reaching term have also been reported and an association with placenta accreta has also been seen (Triolo et al., 2004). Diagnosis can be made in early pregnancy by ultrasonographic visualization of eccentric gestational sac away from midline endometrial echo (Stiller and de Regt, 1991) and rarely by appreciating asymmetrical uterine enlargement on bimanual palpation (Kerr and Anderson, 1934). For definitive diagnosis, laparoscopy is required, where ipsilateral round ligament is seen displaced laterally by the angular gestation as

was seen in our case (arrow in Figure-1a&b). In contrast interstitial pregnancy, which is a close differential diagnosis, displaces the round ligament medially (Jansen and Elliott, 1981). Management of ruptured angular pregnancy includes resuscitation of patient followed by delivery of fetus and repair of the rent or hysterectomy. The delivery of fetus can be executed through either the rent (preferred mode) or via hysterotomy (Stiller and de Regt, 1991). In literature, only four cases of spontaneous mid-trimester rupture of angular pregnancy have been described (Table-1). Three of these cases underwent hysterectomy while one underwent hysterotomy. Ours is the second such case in literature, where spontaneous mid-trimester rupture of angular pregnancy was managed by hysterotomy. In our case, it was not possible to deliver fetus through the rent, so hysterotomy was done. For unruptured angular pregnancy detected on clinical/ ultrasonographic examination, termination of pregnancy should be strongly considered. A meticulous antenatal ultrasonographic evaluation and careful interpretation of features suggestive of angular pregnancy can help not only in preventing a delay in diagnosing this life threatening complication, which if diagnosed before rupture, can be managed by more conservative means.

Conclusion

Though a rare clinical entity, angular pregnancy should be considered as a differential diagnosis in patients presenting with haemoperitoneum in early and mid pregnancy. A knowledge of ultrasonographic features of angular

pregnancy can potentially prevent the occurence of a catastrophic consequence. Declaration of Interest: No work resembling the enclosed article has been published or is being submitted for publication elsewhere. We certify that we have each made a substantial contribution so as to qualify for authorship as detailed at the end of the manuscript. We have not availed of any financial support for this work and do not have any potential conflicts of interest.

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