CASE REPORT

Torsion and rupture of a non-communicating rudimentary horn in a 17-week gestation in a 16-year-old girl: lessons learnt

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Accepted 4 December 2017

SUMMARY

A unicornuate uterus with non-communicating rudimentary horn has always been notorious and poses threat to continuation of pregnancy with dismal consequences. We are reporting an interesting case of uterine malformation with a 90° rotation of uterine axis which ultimately ruptured during termination of pregnancy. The rarity in our case was not only conception in non-communicating horn but also the complete twisting of axis which made the pregnant horn come in front of the non-gravid unicornuate uterus, mimicking normal pregnancy. The most important lesson learnt is that if induction does not lead to cervical changes and uterine contractions, one must consider atypical presentations of an anomalous uterus as a possible differential before proceeding further.

BACKGROUND

A unicornuate uterus with non-communicating rudimentary uterine horn has always been a notorious uterine malformation which threatens normal continuation of pregnancy. The European Society of Human Reproduction and Embryology has revised the classification system of congenital female genital tract anomalies.¹ In light of previous experiences reported in literature, it is well known that a bicornuate or unicornuate uterus with pregnancy in rudimentary horn can lead to uterine rupture, catastrophic haemorrhage and dismal consequences. Despite this, most of the times, diagnosis is made in retrospect when patient has already come with rupture. We are reporting an interesting case in which the diagnosis was missed in the antenatal period and ended in uterine rupture in the second trimester. Laparotomy revealed pregnancy in non-communicating horn of the unicornuate uterus with a 90° rotation of uterus around the longitudinal axis bringing the gravid horn anteriorly. We wish to emphasise that if medical induction of labour does not lead to any cervical change, we must consider atypical presentation of uterine anomalies.

CASE PRESENTATION

A 16-year-old unmarried girl, studying in class 12th, came to us in emergency for termination of 17-week teenage pregnancy. She gave history of having a consensual relationship. Transabdominal ultrasonographic (USG) imaging revealed a dead intrauterine

fetus with well-defined uterine contours and type 2a placenta. Neither there was any breech in uterine contour nor any free fluid.

On examination, the girl was having significant pallor. General and systemic examinations were within normal limits. On per abdominal examination, external ballottement could be elicited in a 16-week gravid uterus. There was no tenderness, guarding or rigidity. Per speculum examination showed a conical cervix with a pinpoint external os. On per vaginal examination, cervix was soft, closed and uneffaced without any discharge or bleeding.

INVESTIGATIONS

Her haemoglobin was 5.5 g/dL. Rest of the blood counts were within normal limits and viral markers were negative. Renal and liver function tests and coagulogram were also normal. Transabdominal ultrasound (US) imaging confirmed a single dead fetus in utero without any suspicion of rupture.

DIFFERENTIAL DIAGNOSIS

The patient had history of menorrhagia and was carrying previous reports of haemoglobin ranging between 7 and 9 g/dL prior to pregnancy. As there was no definitely evident haemorrhage, this was anticipated as the cause of severe anaemia. With a normal 16-week ballottable uterus in midline, initially there was no doubt for diagnosis but when she did not respond to usual methods of second trimester abortion with prostaglandin, some other factor was suspected. The chief differentials were rupture of normal uterine cavity, rupture of a non-communicating rudimentary horn or a cornual ectopic pregnancy.

TREATMENT

The situation was challenging considering the tender age of the patient, dead fetus, feeling of guilt and depression in the patient, the social stigma attached to an unwed mother and family pressure on parents. The patient was transfused 4 units of packed red blood cells to correct anaemia. In view of her being a primigravida, a decision for medical termination of pregnancy was made using mifepristone 200 mg followed 24 hours later by misoprostol, 400 µg at 4 hourly intervals for five doses. There were mild contractions but essentially no change in cervix even after administration of 2000 µg of misoprostol. She was given rest for 24 hours

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To cite: Kumar N, Das V, Pandey A, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-222073

Figure 1 Ruptured uterine horn on the right side with clamps applied on it for resection.

before reevaluating her for extra-amniotic Emscridil instillation but she developed fever (101° Fahrenheit) with tachycardia after 12 hours. As her general condition was fine and she was maintaining her blood pressure, so we continued to give intravenous antibiotic and antipyretics. Subsequently she developed tenderness in lower abdomen with abdominal guarding though there was no bleeding per vaginum. Transabdominal sonography showed dead fetus in the uterine cavity with mild free fluid in abdomen with few heterochoic shadows behind the uterine body which was reconfirmed with transvaginal sonography also. A decision for emergency laparotomy was immediately taken. Preoperatively we found rupture of gravid uterine horn with a normal-sized unicornuate uterus lying posteriorly. The uterine axis had turned 90° bringing the pregnant rudimentary horn anterior and the small unicornuate uterus lying posteriorly. The rudimentary horn was non-communicating and had a shaggy, oedematous fallopian tube. The rudimentary horn was resected

Figure 2 Unicornuate uterus and tube on the left side with attached ruptured uterine horn on the right side.

along with the tube. [Figure 1](#) shows the ruptured uterine horn and [figure 2](#) shows the unicornuate uterus with a normal tube.

OUTCOME AND FOLLOW-UP

The patient had around 150 cc serosanguinous collection in drain over 12 hours which progressively lessened and drain was removed after 48 hours. She had paralytic ileus till 24 hours which was managed conservatively. She did well during the post-operative stay for 7 days and was sent to adolescent counselling centre after being discharged from the hospital.

DISCUSSION

Teenage pregnancy with conception in non-communicating rudimentary horn was possible perhaps due to transperitoneal migration of a sperm or a fertilised ovum which indeed is a rare event. Pregnancy in rudimentary horn is frequently missed and commonly patients present in shock due to rupture of horn. [Thakur *et al*²](#) reported a ruptured rudimentary horn at 19-week pregnancy with previous caesarean section. The diagnosis was missed in a routine anomaly scan and the patient came in shock. A very similar case was reported by [Kanagal and Hanumanalu³](#) where a 25-year-old woman with previous one vaginal delivery had 25-week pregnancy with intrauterine demise. The case was misdiagnosed as intrauterine pregnancy and misoprostol induction was started. She had rupture of non-communicating rudimentary horn of the unicornuate uterus and the patient presented in shock. [Dhar⁴](#) also reported a case of undiagnosed rudimentary horn pregnancy at 22 weeks who presented in shock with features of acute abdomen. Sometimes the uterine rupture has also been reported as late as 31 weeks.⁵ In all of the above patients, rudimentary horn was excised with ipsilateral salpingectomy. Timely diagnosis of this rare mullerian anomaly is possible prenatally but with poor sensitivity. [Tsafir *et al*⁶](#) proposed criteria for US diagnosis which includes the presence of a pseudo pattern of an asymmetrical bicornuate uterus, absent visual continuity of tissue surrounding the gestation sac and the uterine cervix and the presence of myometrial tissue surrounding the gestation sac.⁶ However, the peculiarity in our case was a very rare right-angle turn of the uterus on its longitudinal axis which brought the enlarged rudimentary horn anteriorly towards midline.

The diagnosis of mullerian abnormalities is phenomenally difficult in pregnancy because of distension of pregnant horn. Moreover, the other more reliable tests like hysteroscopy, saline infusion sonography and hysterosalpingography cannot be carried during pregnancy. However, MRI is of great use in view of its safety in pregnancy and high accuracy in soft tissue evaluation. A type of torsion was reported by [Singh *et al*⁷](#) in which MRI was done, in a patient with previous full-term normal vaginal deliveries presenting in this pregnancy at 20 weeks due to pain in abdomen. It revealed an unruptured gravid non-communicating rudimentary horn that was twisted along with the ipsilateral ovary. An important clinical clue to diagnosis again was failure of prostaglandin induction with no cervical changes. This point is also reported by [Abreu *et al*⁸](#) in their case report on two patients who were diagnosed at the time of labour induction following fetal demise. [Samuels and Awonuga⁹](#) reported a young primigravida with a history of an US diagnosis of bicornuate uterus who presented with mild abdominal pain. USG revealed a viable 18-week fetus with anhydramnios in the left uterine horn. Labour induction with misoprostol culminated in uterine rupture. At laparotomy, a ruptured left non-communicating rudimentary uterine horn of a unicornuate uterus was noted. So, failure of prostaglandin

induction to initiate labour and cervical changes could be a hint to the presence of müllerian uterine anomaly. It has to be kept in mind in cases of antepartum fetal death and mid-trimester abdominal pain. Therefore, a high index of suspicion combined with clinical examination and imaging in form of three-dimensional US and MRI are important to establish a correct diagnosis of unruptured rudimentary horn.

Rarely the rudimentary horn is distensible enough to carry the gestation up till near term. Gonçalves *et al*¹⁰ reported a case where incidentally while taking the patient for caesarean due to malpresentation at 34 weeks, a live fetus was found in non-communicating rudimentary horn. Kawthalkar *et al*¹¹ also reported a case with a good neonatal outcome in a case of pregnancy in rudimentary horn. Another important differential to be kept in mind is cornual ectopic pregnancy. Shergill *et al*¹² presented a report when a patient who was taken up for surgery as cornual ectopic had conception in non-communicating rudimentary horn.

The rarity in our case was not only conception in non-communicating horn of the unicornuate uterus but also the complete twisting of the uterus along its longitudinal axis which made the pregnant horn to come in front of the unicornuate uterus, mimicking normal pregnancy. The most important learning

lesson is to stop and revise a checklist and consider these rare conditions as possible differentials before proceeding further.

Contributors NK: did the surgery and wrote the draft. VD: designed the concept, helped in formulating and corrections. SA and AP: helped in literature search.

Competing interests None declared.

Patient consent Guardian consent obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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REFERENCES

- 1 Grimbizis GF, Gordts S, Di Spiezio Sardo A, *et al*. The ESHRE/ESGE consensus on the classification of female genital tract congenital anomalies. *Hum Reprod* 2013;28:2032–44.
- 2 Thakur S, Sood A, Sharma C. Ruptured noncommunicating rudimentary horn pregnancy at 19 weeks with previous cesarean delivery: a case report. *Case Rep Obstet Gynecol* 2012;2012:1–4.
- 3 Kanagal DV, Hanumanalu LC. Ruptured rudimentary horn pregnancy at 25 weeks with previous vaginal delivery: a case report. *Case Rep Obstet Gynecol* 2012. doi: 10.1155/2012/985076. [Epub ahead of print 6 Jun 2012].
- 4 Dhar H. Ruptured rudimentary horn at 22 weeks. *Niger Med J* 2012;53:175–7.
- 5 Al Qarni AA, Al-Braikan N, Al-Hanbali MM, *et al*. Rupture rudimentary horn pregnancy at 31 week. *Saudi Med J* 2017;38:201–3.
- 6 Tsafirir A, Rojansky N, Sela HY, *et al*. Rudimentary horn pregnancy: first-trimester prerupture sonographic diagnosis and confirmation by magnetic resonance imaging. *J Ultrasound Med* 2005;24:219–23.
- 7 Singh P, Gupta R, Das B, *et al*. Midtrimester spontaneous torsion of unruptured gravid rudimentary horn: Presurgical diagnosis on magnetic resonance imaging. *J Obstet Gynaecol Res* 2015;41:1478–82.
- 8 Abreu R, Barros S, Jardim O, *et al*. Failure of prostaglandin induction labor in a Müllerian abnormality. *Arch Gynecol Obstet* 2010;282:143–7.
- 9 Samuels TA, Awonuga A. Second-trimester rudimentary uterine horn pregnancy: rupture after labor induction with misoprostol. *Obstet Gynecol* 2005;106:1160–2.
- 10 Gonçalves E, Prata JP, Ferreira S, *et al*. An unexpected near term pregnancy in a rudimentary uterine horn. *Case Rep Obstet Gynecol* 2013;2013:1–4.
- 11 Kawthalkar AS, Gawande MS, Jain SH, *et al*. Rare case of live birth in a ruptured rudimentary horn pregnancy. *J Obstet Gynaecol Res* 2011;37:1169–72.
- 12 Shergill HK, Suparna G, Chhabra A. Rare presentation of ruptured rudimentary horn pregnancy. *Int J Med Res & Health Sci* 2015;4:925–7.

Learning points

- ▶ Unicornuate uterus with non-communicating rudimentary horn is a rare congenital uterine malformation and is known to result in adverse pregnancy outcomes.
- ▶ One has to keep in mind this rare finding before planning termination.
- ▶ 90° torsion of the axis can masquerade as normal uterine body and no cervical change even with labour-inducing agents can be a clue to early diagnosis.

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