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Case Report

Pregnancy in a uterine anomaly: a case report

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ABSTRACT

Pregnancy in rudimentary horn of the uterus is a rare clinical condition and often misdiagnosed as tubal, cornual, abdominal and even intrauterine pregnancy. Surgical excision of the horn is the definitive treatment for this condition. A healthy primigravida at 11 weeks gestation with underlying corrected Tetralogy of Fallot was seen with ultrasound findings suggestive of ectopic pregnancy. She underwent laparotomy and intraoperative finding was unruptured right non-communicating rudimentary horn pregnancy which was surgically excised. This case highlights the importance of having this rare diagnosis as one of the differential diagnosis for ectopic pregnancy.

Keywords: Rudimentary horn, Surgical excision, Tetralogy of Fallot

INTRODUCTION

Unicornuate uterus with rudimentary horn is an anomaly resulting from partial development of one of the Müllerian ducts and an incomplete fusion with its contralateral side. Unicornuate uterus with rudimentary horn is a rare condition affecting 1:10000 to 1:140000 pregnancies.[1] Rudimentary horn pregnancies usually present with acute abdomen when the horn ruptures with advancing gestation. This condition could be dangerous as it may cause significant maternal morbidity and even mortality. In Malaysia, one case was reported in 2011 where the diagnosis was made intra-operatively when patient presented with acute abdomen with hypovolemic shock.[2] Our case was clinically stable and the rudimentary horn pregnancy was only diagnosed intra-operatively.

CASE REPORT

24 year old healthy primigravida at 11 weeks pregnancy with background history of corrected Fallot's Tetralogy was referred from health clinic for molar pregnancy. Clinically she was pink and her vital signs were stable. Abdomen was soft, non tender and the uterus was not palpable. Vaginal examination revealed a single cervix with a closed cervical os.

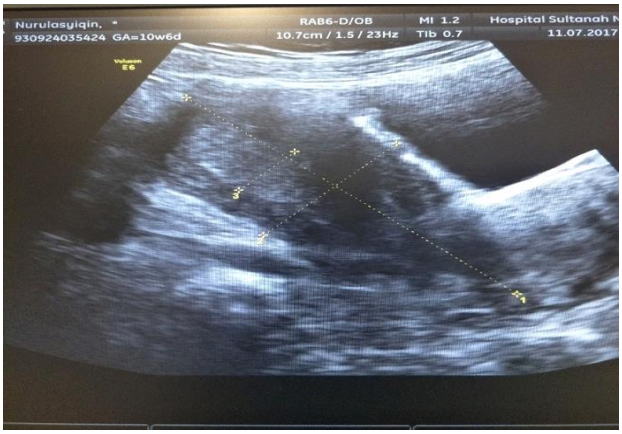

Figure 1: Empty uterus with thick endometrium.

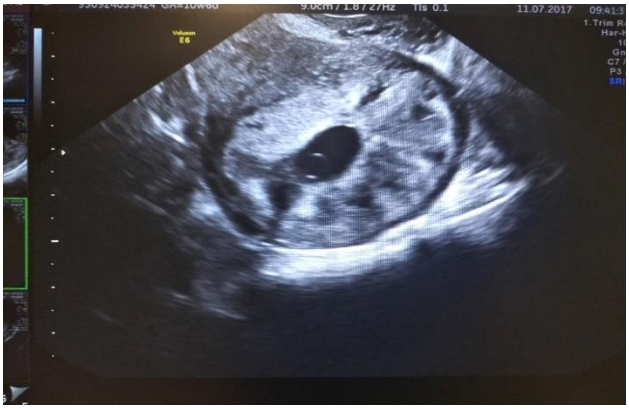

Figure 2: Transvaginal scan showed gestational sac and yolk sac encapsulated by mass with myometrium-tissue like.

Pelvic ultrasound showed an empty uterus with thick endometrium (Figure 1). There was a right adnexal mass measuring 5x6cm with a central double trophoblastic ring containing a yolk sac (Figure 2). No obvious foetal echo was visualised. Myometrial-like tissue was also seen surrounding some parts of the adnexal mass. The mass however was seen away and separated from the uterus. Despite this finding, diagnosis of unruptured right cornual ectopic pregnancy was made.

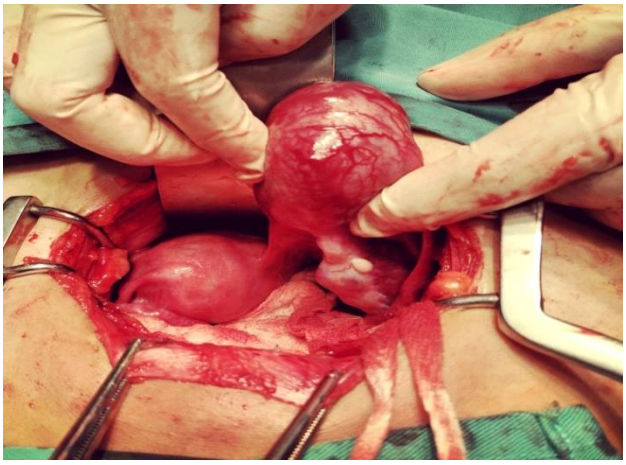

Figure 3: Right rudimentary horn with bilateral.

She underwent exploratory laparotomy under general anaesthesia. Intraoperative findings were unruptured right rudimentary horn pregnancy (Figure 3).

Left unicornuate uterus was otherwise normal. There was a fibrous band between the right horn and the left uterus but direct communication between these two structures was absent. Both ovaries and tubes were normal. Excision of the right rudimentary horn was performed and cut section showed myometrium and gestational sac (Figure 4).

Post-operative period was uneventful, and she was discharged well few days operation.

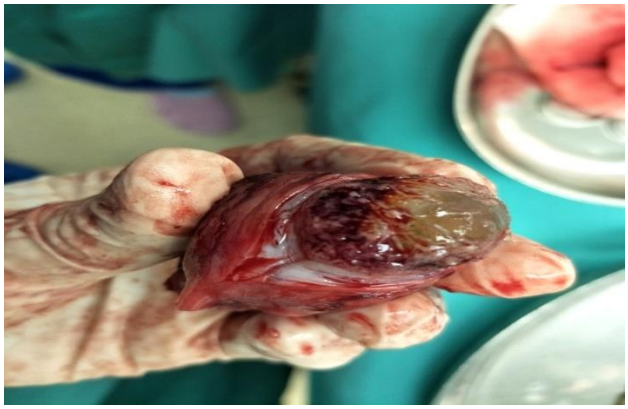

Figure 4: Cut section showed gestational sac.

DISCUSSION

Unicornuate uterus with rudimentary horn is a rare condition affecting 1:10000 to 1:140000 pregnancies.[1] It is classified as class II ASRM (American Society of Reproductive Medicine) type of Mullerian Anomaly. It could be further divided into 4 types including:

- Cavitary communicating rudimentary horn
- Cavitary non-communicating rudimentary horn
- Non cavitary rudimentary horn
- Absent rudimentary horn.

Pregnancy in rudimentary horn is extremely rare. Information regarding this condition is based mainly on literature case reports. Although seemingly impossible, pregnancy in non-communicating rudimentary horn has been reported and interestingly it actually comprised of the majority of reported cases reported in the literature.[3]

Trans-peritoneal migration of the spermatozoa was quoted in many papers as the possible mechanism for the occurrence of pregnancy. This theory however cannot explain the 10% of cases whereby the corpus luteum was observed on the contra-lateral side. It is probable that in such cases, fertilization occurs in the peritoneal cavity with subsequent transmigration and transplantation of the fertilized ovum in the rudimentary horn. Latto and Norman in their classic paper published in 1950 on the other hand believe that the theory of trans-peritoneal migration is untenable based on few sound arguments.[4] They believe that pregnancy invariably occurs in communicating rudimentary horn but manifested eventually as non-communicating rudimentary horn pregnancy because tissues reacting to the advancing syncytium in pregnancy will cause occlusion of the communication channel.

Early detection of rudimentary horn pregnancy is crucial to avoid high morbidity and mortality risk. Early detection however is not easy unless there is a high index of suspicion since the sensitivity of ultrasound is only 26%.[5] Tsafrir et al suggests several criteria including:

- a pseudo pattern of asymmetrical of bicornuate uterus;
- absent visual continuity tissue surrounding the gestation sac/ uterine cervix; and
- presence of myometrial tissue surrounding the gestation sac; to suggest pregnancy in the rudimentary horn.[6]

As the pregnancy advances, the sensitivity of ultrasound becomes a lot less. Most cases are in fact diagnosed intraoperatively in the 2nd trimester when rupture occurs. Few reported cases advanced to the 3rd trimester.[3,7] Out of the 10% of cases that reach term, and the fetal salvage rate is only 2%.[8] Rudimentary horn pregnancy was often misdiagnosed as tubal, cornual, abdominal and even intrauterine pregnancy.[9,10] Preoperatively, we diagnosed this case as cornual pregnancy since myometrial tissue was seen surrounding the gestational sac. This is despite documenting that the 'ectopic gestation' was not continuous with the empty uterus.

The aetiology behind rupture of the rudimentary horn in pregnancy is due to the underdevelopment of the myometrium. The onset of rupture largely depends on the variable thickness of musculature in addition to the dysfunctional endometrium.[11] In addition, poorly developed musculature can also cause placenta percreta and the reported incidence was quoted to be around 11.9%.[8] Placenta percreta can be confirmed by a histopathology examination from as early as seven weeks.[12]

Surgical excision is the definitive treatment of rudimentary horn pregnancy in order to prevent complication such as rupture, recurrence and chronic pelvic pain.[13,14] The choice of surgical approach either via laparoscopy or laparotomy depends on patient's general condition, gestational age, size and vascularity of the pregnant horn. First trimester diagnosis facilitates laparosocopic management with rapid favourable outcome while laparotomy on the other hand is usually reserved for patients with acute abdomen.[6,15]

Some authors reported the successful use of systemic methotrexate administration; however this approach does not prevent recurrence.[13] Park et al described combination of both medical and surgical management of rudimentary horn pregnancy. In his paper, feticide with intracardiac potassium chloride and intraplacental methotrexate were given to reduce blood flow to facilitate subsequent interval laparoscopic excision.[16] However, in our case, although diagnosis of ectopic was made at early gestation, laparoscopy was not performed due to her underlying cardiac condition.

CONCLUSION

Rudimentary horn pregnancy is a rare phenomenon; however this diagnosis should be entertained when the diagnosis of ectopic pregnancy is made. The purpose of this report is to create awareness amongst clinicians of this very rare diagnosis.

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