Case Report

Multiple Faces of the Same Pathology

Ng KS[1], Fazarina M[2], Anizah A[1], Shuhaila A[1], Yulianty A[1](✉), Zainul Rashid MR[1]

[1]Department of Obstetric and Gynaecology, [2]Department of Pathology, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, Bandar Tun Razak, 56000 Cheras, Kuala Lumpur, Malaysia.

Abstract

Ectopic pregnancy is defined as an extrauterine pregnancy. We report three cases where the ectopic pregnancies were implanted in different sites. The first case was a 28-year-old in her second pregnancy at 9 weeks gestation. She presented with painless vaginal bleeding. Ultrasound showed unruptured cornual pregnancy with hCG level of 7456mIU/ml. A single dose of 75mg IM methorexate was given and she responded well with significant reduction of hCG level. The second case, a 26-year-old gravida 5 para 2+2, with 2 previous ectopic pregnancies and bilateral salpingectomy, conceived via in-vitro fertilization (IVF). She presented with acute abdomen and one episode of syncope at 8 weeks 4 days gestation. Laparotomy showed ruptured ectopic pregnancy at the left tubal stump requiring a left salpingectomy. The third case was a 26-year-old, gravida 5 para 2+2, with two previous vaginal deliveries and two previous first trimester miscarriages. Her menses was irregular since she took injectable progestin. She presented to the emergency department with sudden onset of lower abdomen pain. Urine pregnancy test was positive. Ultrasound showed empty uterus, no adnexal mass but there was significant free fluid in the cul-de-sac. During laparoscopy, a ruptured ovarian pregnancy was diagnosed and salpingo-oophorectomy performed. There was no significant risk factor contributing to ectopic pregnancy identified in the first and third case. In the second case, despite previous bilateral salpingectomy, the patient still had ectopic pregnancy in the left fallopian tube remnant.

Keywords: Cornual pregnancy, laparotomy, methotrexate, ovarian pregnancy, salpingectomy, tubal stump pregnancy

Correspondence:

YuliantyArifuddin, Department of Obstetric and Gynaecology, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Jalan Yaacob Latif, Bandar Tun Razak, 56000 Cheras, Kuala Lumpur, Malaysia. Tel: +603-91455949 Fax: +603-91456672 Email: yuliantya77@gmail.com

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Introduction

The incidence of ectopic pregnancies has increased significantly due to increased rate of pelvic inflammatory disease and ART procedures. About 98% occur in the fallopian tube. Other possible sites include interstitial (cornual), caesarean scar, ovarian and abdominal.We report three rare different sites of ectopic pregnancy.

Case Report

Case Report 1 - Cornual Pregnancy

A 28-year-old, gravida 2 para 1, at 9+ week gestation following spontaneous conception, presented with 3 days history of painless per vaginal bleeding. She did not have anaemic symptoms and denied passing out product of conception.

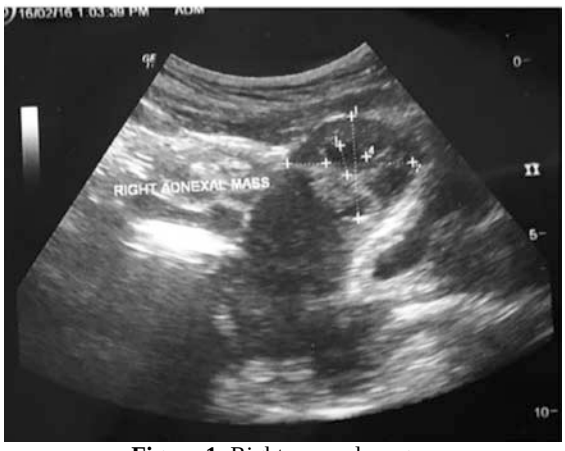

Figure 1: Right cornual pregnancy

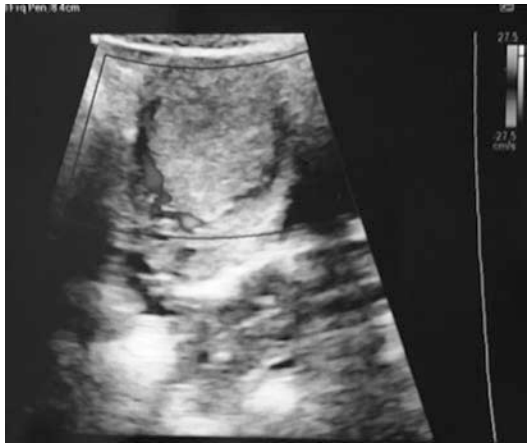

Figure 2: Doppler scan of cornual pregnancy

She had a previous vacuum assisted delivery for fetal distress with no history of intrauterine contraceptive device (IUD) insertion, pelvic inflammatory disease or endometriosis.

She was hemodynamically stable and abdominal examination was unremarkable. Speculum examination revealed minimal blood. Vaginal examination revealed a 6 weeks anteverted, mobile uterus, closed cervical os and absence of cervical tenderness. No adnexal mass was palpable.

Transvaginal ultrasonography showed an empty uterus. An ectopic mass, measuring 3.1x2.8cm was seen at the right uterine cornu (Fig. 1) with presence of 'ring of fire' around the mass on Doppler imaging (Fig. 2). No free fluid was seen in the pouch of Douglas. Quantitative β-hCG was 7456 mIU/mL.

She was given a single dose of 75mg (50mg/m2) intramuscular methotrexate. She remained stable with a significant reduction in beta HCG level to 1248mIU/ml at 14th day post treatment.

Case Report 2 – Tubal Stump Pregnancy

A 26-year-old, gravida 5 para 2+2, with 2 previous normal vaginal deliveries followed by 2 ectopic pregnancies requiring a laparoscopic right salpingectomy and left salpingectomy via laparotomy respectively in 2011.

She conceived by in-vitro fertilization in the fifth pregnancy. An ectopic pregnancy was suspected at 5 weeks post embryo transfer. She requested conservative treatment but presented at 8+ weeks gestation with an acute abdomen with one episode of syncope. She had minimal per vaginal bleeding.

On examination, she was in pain, pale and hypotensive. Her abdomen was distended, guarded and tender over the lower abdomen. Per speculum examination revealed a healthy looking cervix with minimal blood. Vaginal examination revealed a closed cervical os, a normal-sized uterus and fullness at the left adnexa. Cervical excitation was positive.

Ultrasound scan demonstrated an empty uterus with a left adnexal mass, measuring 3.5 x 2.6 cm. Free fluid was seen in the pouch of Douglas. Her hemoglobin was 6.0 g/dl.

An emergency laparotomy and resection of left tubal stump was performed, revealing a 1.6 liter hemoperitoneum and a ruptured ectopic mass (3x3cm) at the left tubal stump. The right fallopian tube was absent. The uterus was normal and hemorrhagic cysts were seen in both ovaries.

She required three pints of packed cell blood and 2 units of fresh frozen plasma. Histological examination confirmed a left tubal pregnancy.

Case Report 3 – Ovarian Pregnancy

A 26-year-old, gravida 5 para 2+2, presented with sudden onset of lower abdominal pain for 1 day. Her menstrual cycle was irregular and was amenorrhoeic for the past 3 months. She had two normal vaginal deliveries and two previous first trimester miscarriages, one of which required suction and curettage. This was a spontaneous conception.

She did not have any vaginal bleeding or syncope but was pale and hypotensive with hemoglobin level of 11.6 g/dl. Abdominal examination revealed tenderness over lower abdomen with guarding. Vaginal

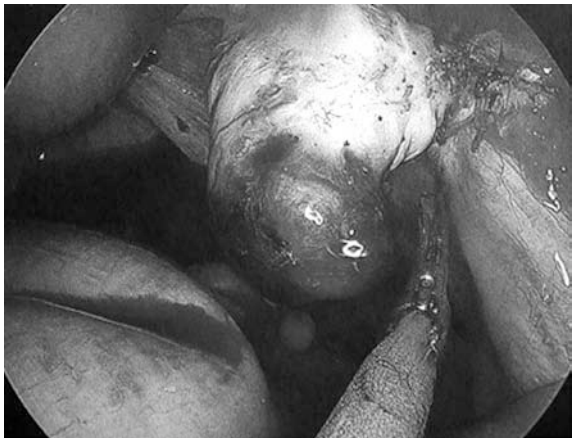

Figure 3: Ovarian pregnancy

examination showed a normal size uterus, closed cervical os and tenderness at right adnexae. On ultrasonography, the uterus was empty with no adnexal mass and normal ovaries. However, significant free fluid was seen in the cul-de-sac.

An emergency diagnostic laparoscopy was performed for suspected ruptured right tubal ectopic pregnancy. The right ovary was enlarged with a ruptured ovarian ectopic, measuring 2 x 3cm (Fig 3). There was 1.7 liter of hemoperitoneumn requiring 2 units packed cell transfusion. The uterus, both fallopian tubes and left ovary were normal. Right salphingo-oophorectomy was performed.

Histopathological examination confirmed a ruptured right ovarian ectopic pregnancy.

Discussion

The three cases of ectopic pregnancy involved different extra-uterine location with various symptoms and signs.

Cornual (interstitial) pregnancy is a rare type of ectopic pregnancy, accounting for 2-4% of all tubal pregnancies. The risk factors are history of previous ectopic pregnancy, pelvic inflammatory disease, assisted reproductive conceptions as well as tubal surgery (salpingectomy and salpingostomy) (1,2), which were all absent in this patient. The diagnosis of cornual pregnancy is usually late due to good muscular and vascular support of this part of fallopian tube allowing good distensibility of ectopic mass, thus causing less pain. By the time it ruptures at advanced gestation, it may result in catastrophic haemorrhage with high mortality rate of 2% (3). Fortunately, she presented early at 9 weeks gestation, allowing early detection, intervention and prevention of fatal massive haemorrhage. In experienced hands, transvaginal ultrasound can establish the diagnosis in nearly 71% of cases (4) which can be further improved using these criteria: an empty uterus, gestational sac seen separately and <1 cm from the most lateral edge of the uterine cavity and a thin myometrial layer surrounding the sac (5). In our patient, the first two criteria were present with an additional Doppler study showing vasculature 'ring of fire' around the mass at uterine cornua. Surgical interventions include laparotomy or laparoscopic cornual resection. Medical treatment involved the use of methotrexate, injected at the ectopic site or systemically as a single intramuscular injection. Medical treatment is only advocated for those who are hemodynamically stable, beta subunit of human chorionic gonadotropin (HCG) value <5000 mIU/mL, absent fetal heart activity, anadnexal mass of ≤4 cm and hemoperitoneum of <100 mL (6). Our patient fulfilled most of these criteria and was given intramuscular injection. She responded well with significant reduction of β-hCG levels without any complication.

The case of tubal stump pregnancy is really a mishap. Nevertheless, in combination with a history of two previous ectopic pregnancies and an in-vitro fertilization conception, which are significant risk factors, this favoured a repeat ectopic pregnancy. The rate of ectopic pregnancy following ART is 1.7%-2% of all pregnancies (7) and it is rarely seen at the proximal tubal stump among patients who had bilateral salpingectomy. Isthmic ectopic pregnancy carries a mortality rate of around 2.0–2.5%, as compared with other ectopic pregnancies of only around 0.14 % (8).

This patient was suspected to have an asymptomatic ectopic pregnancy as early as 5 weeks POA following IVF but she opted for conservative management. Due to the poor distensibility and high vascularity of this portion of the Fallopian tube, it is associated with high risk of rupture and severe hemorrhage at an early gestation (9) as had happened in this patient who presented at 8 weeks gestation with acute abdomen and hypovolemic shock requiring an emergency laparotomy.

Despite bilateral salpingectomy, ectopic pregnancies can still occur. Thus, to reduce the incidence, total salpingectomy (instead of partial salpingectomy) and cauterization of the tubal stump (10) should be advocated. Marcus & Brinsden (11) suggested that post-ART ectopic pregnancies can originate either from the direct transfer of embryo to the tube or migration of the embryo from the endometrial cavity to the tube. Embryo transfer technique (deep fundal transfer) without visual assistance, use of high volume transfer medium, multiple embryo transfer, frozen

embryo transfer and artificial insemination can all increase the possibility of ectopic pregnancies (11).

Ovarian pregnancy is another rare variant of ectopic pregnancy. Hertig estimated that it occurs in one in 25,000-40,000 pregnancies (12). Intrauterine device usage was found to have a strong association with ovarian pregnancy (13). It is characterized by a poor clinical symptomatology and a difficult ultrasound diagnosis as the gestational sac may resemble a corpus luteum, a hemorrhagic cyst or an endometriotic cyst of ovary. Hallat (14) study showed out of 25 cases of suspected ovarian pregnancy, only 28% was the correct diagnosis at surgery. It strongly suggests that distinguishing between ovarian pregnancy and corpus luteal/hemorrhagic cyst rather difficult. In this patient, ultrasonography findings showed an empty uterus, no adnexal mass visualized but with a significant free fluid in the cul-de-sac. There was no risk factor identified in our patient such as previous IUD usage and the diagnosis was missed on ultrasonography. Nonetheless, there was no delay in surgery as the diagnosis of ruptured ectopic pregnancy was obvious. Surgical treatment can be either wedge resection or oophorectomy (15). In this patient, an oophorectomy was performed. Medical management remains debatable. However, it can be carried out in patients who fulfill the criteria based on Greentop Guidelines (6). As mentioned, diagnosis is difficult to be ascertained. If a diagnostic laparoscopy was performed, treatment should be initiated concurrently.

Generally, treatment options are dependable on integrity of ectopic mass and hemodynamic stability of patient during presentation. It is essential to have a proper patient selection, thorough counseling and informed consent prior to medical intervention. The last two cases involved unstable patients with ruptured ectopic pregnancies requiring life-saving surgical intervention. Tubal stump pregnancy remained a rare site for ectopic. Thus, total salpingectomy, cauterization of the stump and avoid tubal clipping should be advocated, particularly in women undergoing IVF. Ovarian pregnancy remains a challenge to all obstetricians to obtain a correct diagnosis before medical intervention.

Conclusion

Ectopic pregnancy, disregard of its implantation site, is a potentially life-threatening condition. An early diagnosis can facilitate timely intervention especially with less invasive treatment and avoid major complications, hence reduce maternal morbidity and mortality.

References

1. Tulandi T, Al-Jaroudi D. Interstitial pregnancy: results generated from the Society of Reproductive Surgeons Registry. Obstet Gynecol 2004; 103(1): 47-50.

2. Agarwal SK, Wisot AL, Garzo G, Meldrum DR. Cornual pregnancies in patients with prior salpingectomy undergoing in vitro fertilization and embryo transfer. Fertil Steril 1996; 65(3): 659-60.

3. Alam IP. Cornual pregnancy: a case report. Faridpur Med Coll J 2013; 8(2): 102-4.

4. Abraham D, Silkowski C. Emergency Medicine Sonography 1st ed; 2010, 264-27.

5. Timor-Tritsch IE, Monteagudo A, Matera C, Veit CR. Sonographic evolution of cornual pregnancies treated without surgery. Obstet Gynecol 1992; 79(6): 1044-9.

6. Royal College of Obstetricians and Gynaecologists (RCOG). The Management of Tubal Pregnancy. Clinical Guideline No. 21. London: RCOG Press; 2004. Reviewed 2010:1-10.

7. Society for Assisted Reproductive Technology: American Society for Reproductive Medicine. Assisted reproductive technology in the United States: 2001 results generated from the American Society for Reproductive Medicine/Society for Assisted Reproductive Technology registry FertilSteril 2007; 87(6): 1253-66.

8. Lau S, Tulandi T. Conservative medical and surgical management of interstitial ectopic pregnancy. Fertil Steril 1999; 72(2): 207-15.

9. Takeda A, Manabe S, Mitsui T, Nakamura H. Spontaneous ectopic pregnancy occurring in the isthmic portion of the remnant tube after ipsilateraladnexectomy: report of two cases. J Obstet Gynaecol Res 2006; 32(2): 190-4.

10. Garavaglia E, Quaranta L, Redaelli A, Colombo G, Pasi F, Candiani M. Interstitial Pregnancy after In Vitro Fertilization and Embryo Transfer Following Bilateral Salpingectomy: Report of Two Cases and Literature Review. Int J Fertil Steril 2012; 6(2): 131-4.

https://doi.org/10.17576/JSA.2016.0602.13

11. Marcus SF, Brinsden PR. Analysis of incidence and risk factors associated with ectopic pregnancy following in-vitro fertilization and embryo transfer. Hum Reprod 1995; 10(1): 190–203.

12. Gerin-Lojoie L. Ovarian pregnancy. Am J Obstet Gynecol 1951; 62(4): 920.

13. Marret H, Hamamah S, Alonso AM, Pierre F. Case report and review of the literature: primary twin ovarian pregnancy. Human Reprod 1997; 12(8): 1813-15.

14. Hallat JG. Primary ovarian pregnancy a report of twenty-five cases. Am J Obstet Gynecol 1982; 143(1): 50-60.

15. Patel Y, Wanyonyi SZ, Rana SF. Laparascopic management of an ovarian ectopic pregnancy: case report. East Afr Med J 2008; 85(4): 201–4.