



DIAGNOSTIC DILEMMA AND THERAPEUTIC CHALLENGES IN MANAGING RARE CASES OF ECTOPIC PREGNANCIES.

Dr Samiha Samad*

*Department of Obstetrics & Gynaecology, Sohar Hospital, Sohar/Oman

*Corresponding Author: Dr Samiha Samad,
*Email: samiha_javed@yahoo.co.uk

Objective: to highlight the difficulties and challenges faced in the management of rare cases of ectopic pregnancy and find the most effective management in our setup.

We present case series of rare cases of ectopic pregnancies managed in SOHAR Hospital. Incidence of rare site was 3.2%.

Introduction

Ectopic Pregnancy (EP) is defined as the implantation of the fertilized egg occurs outside the uterine cavity. Reported incidence is approximately 2% of all pregnancies.⁽¹⁾ The fallopian tube is the most common location, accounting for more than 90% of cases.⁽²⁾ However implantation in the rare sites such as in abdomen (1%), cervix (1%), ovary (1-3%) and caesarean scar (1-3%) can occur and often results in greater morbidity because of delayed diagnosis and treatment.⁽²⁾

We performed 3-year retrospective audit within our department from 09/6/2021-09/06/2024 to review the management outcome of ectopic pregnancy.

408 cases of ectopic pregnancies were managed during this period. Among them 80% were in fallopian tube, remaining were PUL 17% (71/408), scar 0.98% (4/408), cornual 0.73% (3/408), heterotopic 0.49% (2/408), ovarian 0.24% (1/408), Interstitial 0.24% (1/408). Secondary abdominal ectopic was 0.49% (2/408).

As the number of cases of rare presentation of ectopic are very less, most of the gynecologists have either none or limited experience of managing such cases. Therefore, it is important to share our experience of managing these atypical cases and discuss the challenges and ways to deal more effectively and efficiently to minimize maternal morbidity and mortality due to ectopic pregnancy in unusual sites.

Case 1

29-year-old female, G3P2, 6 weeks pregnant with 2 previous LSCS, referred from local health center with complaint of diffuse abdominal pain and vomiting for one day of duration.

On general examination she was conscious well oriented, pale looking with BP 84/44 mm of Hg, pulse was 120/minute, 36C and oxygen saturation was 100%. Abdominal examination confirmed soft and tender with no rebound or rigidity. Complete blood count suggested Hb 10.73g/dl, hematocrit 32.93, WBC 19 LFT, RFT, coagulation profile was normal. Serum β hCG was 2293IU/ml.

Transabdominal and Transvaginal ultrasound scan suggested uterus was normal size and uterine cavity was empty. Both ovaries could not be evaluated separately. Mild to moderate free fluid in the abdomen was present. A large irregular echogenic area was seen in lower abdomen measuring 10x5.5 cm.

Most likely diagnosis was to have ruptured ectopic pregnancy therefore she was taken to theatre. During laparotomy approximately 1-liter of hemoperitoneum and blood clots were removed. Uterus exteriorized as was adherent to the anterior abdominal wall which appeared normal along with both tubes. Left ovary was seen oozing along with the products of conception at the site of probable ectopic mass. Right ovary had 3cm corpus luteum cyst which was also oozing.

(Fig 1): Showing Right ovarian corpus luteal cyst & left ovary with ectopic pregnancy

Fig (2) Showing Specimen of ovarian cyst and ectopic mass

The product of conception removed from the left ovary and bleeding corpus luteal cyst from right ovary. Remaining ovarian tissue was conserved and bleeding was secured with the hemostatic suture.

Histopathology examination confirmed products of conception.

The patient was discharged home in stable condition on 3rd postop day with follow up appointment in outpatient department.

TABLE 1

Beta Hcg	
25/2/2023	2293 mIU/ml (pre op)
26/2/2023	879 mIU/ml (post op)
28/2/2023	270 mIU/ml (post op)

CASE 2

A 33-year-old Omani female, G5P4+1, 11wk pregnant with previous 3 LSCS (1 set of twins) presented in emergency with complains of bleeding PV, with passage of clots. Her LMP was 5/9/23 with EDD 12/6/2023.

On examination the woman was conscious, well oriented and vitally stable. Abdominal examination revealed soft and nontender. Vaginal examination suggested closed cervical os and no active bleeding noted.

Ultrasound scan suggested thickened endometrium of 23 mm and the diagnosis of incomplete miscarriage was suggested. The plan was to admit the patient for medical termination with Misoprostol as per protocol.

The patient received 4 doses of Misoprostol with no expulsion of product of conception or no bleeding per vaginum.

Repeat USG was performed to confirm retained products of conception and patient was kept nil by mouth for surgical evacuation if deemed necessary.

Transabdominal ultrasound scan suggested GS with fetal pole (no cardiac activity seen), bulging into the bladder suggestive of scar pregnancy. Therefore, transvaginal ultrasound scan was carried out which also was suggestive of scar pregnancy. The findings confirmed empty endometrial cavity (endometrial thickness of 0.5 mm) with intrauterine gestational sac (1.8x1.6 cm with fetal pole) equivalent to 6weeks and 3 days, with no cardiac activity, embedded in the myometrium in the lower uterine segment, bulging into the bladder.

Fig 3: USG showing scar pregnancy

Diagnosis and options of treatment was discussed with the couple including surgical and medical treatment with all risks and benefits. Couple opted for medical treatment with methotrexate and accepted the risk of long follow up, need of repeat injection and the urgent laparotomy/hysterectomy in case of uncontrolled bleeding.

Methotrexate Injection was given after taking informed consent.

Patient was followed in outpatient department with serial β Hcg

Table 2

Serum Beta Hcg	before methotrexate.
25/11/23	4439iu/ml
	After methotrexate.
2/12/23	1053 IU/ml
6/12/23	61IU/ml
13/12/23	22.70 IU/ml

Case 3

36-year-old, G4P1+2, 8 weeks pregnant, unbooked case referred from local health center (Khasab) because of suspected heterotopic pregnancy.

Her LMP was 5/3/2023 with Irregular cycles. She had a history of suprapubic pain and backache with dysuria for 1 day. She had no bleeding or abnormal vaginal discharge and any medical ailments. She was admitted in Khasab health center for pain and received IV fluids, analgesia and antibiotics. Scan showed intrauterine gestational sac with CRL 7wk 3days with right sided complex mass with suspicion of abscess/? heterotopic pregnancy.

Past Obstetrical History

Her 1st pregnancy ended in SVD at term 8 years back, and in 2nd pregnancy had left side ectopic for which she had salpingectomy in 2015 done abroad (Thailand).

3rd pregnancy was a spontaneous conception with heterotopic pregnancy (with left ectopic and salpingectomy was done). Intrauterine pregnancy aborted spontaneously.

4th pregnancy is the current pregnancy.

On general examination vitals were stable with BP 117/64 mm of Hg and pulse 76/min. Abdominal examination was soft and non-tender.

Ultrasound findings suggested intrauterine gestation sac seen which corresponded to 7wk 3 days, with positive FH. Right side tube was dilated with adjacent mass of 2x2 cm with minimal free fluid in the pelvic cavity.

Impression was of Ruptured hemorrhagic corpus luteum cyst and right sided ectopic pregnancy.

Plan was to admit the patient for observation as she was asymptomatic.

Routine laboratory investigations were sent and repeat ultrasound scan was suggested by the senior obstetrician.

Investigations suggested Hb 12.3, Hct 37, WBC 8.92/ul and β Hcg 56833.00 IU/ml. LFT's, RFTs were all normal. Urine culture revealed no growth.

Repeated USG suggested intrauterine gestational sac in the fundus with clearly visible cardiac activity. Diagnosis of viable intrauterine viable pregnancy corresponding to 7 weeks with right corpus luteum cyst was confirmed.

Right ovary was bulky with corpus luteal cyst seen. Left ovary was normal. There was no tenderness noted during the entire length of scanning.

Findings were explained to the couple and patient sent back to local health center for conservative management and regular follow up for pregnancy locally. Patient was admitted on 1/5/2023 and discharged on 2/5/2023 next day.

She was attended emergency department after 4 days with severe RIF pain and minimal PV spotting. On Examination she was vitally stable. Abdominal examination was soft with tenderness in RIF.

USG done suggested intrauterine gestational sac, CRL equivalent to 8 wks, cardiac activity present, minimal fluid in pouch of Douglas. There was no corpus luteum cyst seen.

Patient was kept NPO. Surgical Referral was made to exclude appendicitis. Abdominal USG was arranged by Radiology Department.

Patient was prepared for laparoscopy for the definitive diagnosis and further management.

Laparoscopy Findings confirmed gravid uterus approximately 8wks. Left fallopian tube was absent as had been previously removed. Both ovaries enlarged looking hyper stimulated with multiple follicles.

Right sided tubal heterotopic pregnancy involving the whole of the right tube, seated on the top of right ovary with few blood clots surrounding it and leaking from the fimbrial end was noted. About 150 ml of blood and clots was present. Right Salpingectomy done. Final Diagnosis of right heterotopic pregnancy was confirmed with histopathology examination.

Fig 4- Right tube distorted by ectopic

Patient was stable post operatively and discharged home on 3rd day.

USG repeated at the time of discharge which suggested approximately 9 weeks viable pregnancy.

Fig 5- live pregnancy shown in 3D

Case 4

30 year P2, IUCD in situ admitted with pelvic pain and irregular spotting. β hcg was 2050 mIU/ml. USG showed IUCD in situ with no gestational sac seen, therefore the diagnosis of ectopic or PUL was made.

IUCD was removed after discussing with the patient. The scan and β Hcg was repeated after 48 hours which had increased to 3181IU/ml. TVS showed small gestational sac at the cornual end of left fallopian tube and right corpus luteum cyst Patient was taken to theatre for diagnostic laparoscopy.

Fig 6. TVS showing small gestational sac at the cornual end and right corpus luteum cyst

The findings during diagnostic laparoscopy suggested normal size uterus, small fleshy tissue anteriorly on uterus below the left cornua which was excised and sent for histopathology. The raw area about 2x2 cm left behind over the uterus cauterize. Both fallopian tubes and left ovary were normal. Right ovary noted to have corpus luteal cyst. Post-operative diagnosis of tubal abortion was made. Beta hcg done on 1st post op day had dropped to 1220 IU/ml.

Plan was to discharge the woman home with serial beta hcg till became negative.

She was seen after a week in the outpatient clinic, when she was asymptomatic with repeat beta Hcg 632 IU/ml. Histopathology confirmed product of conception.

On 1/9/2023 presented again with severe abdominal pain and hypovolemic shock. On examination she was cold and clammy; pulse was 120/min, BP 130/93 mm of Hg. On abdomen examination it was distended, tender+++. USG performed suggested uterus with thin ET with free fluid +++ in the pelvis and abdomen. Laboratory investigation suggested Hb 4 gm%, HCT 12, WBC 18000, platelets were 168, and coagulation screen suggested INR 1.48 and APTT 24.4.

Emergency laparotomy was carried out which confirmed massive hemoperitoneum. Right fallopian tube and both ovaries looked normal. Left fallopian tube was mildly edematous, palpated as normal. A 2cm raw area oozing just below the left cornual end on the anterior surface of uterus sutured. The left broad ligament looked boggy?clots. Vascular surgeon was called who explored retroperitoneum and found small clots and marked congestion and ruled out major vascular or organ injury. 6 units PRBC, 6FFP, 4 platelets were transfused. Intraoperative inotropes was given and patient transferred to ICU in stable condition. As no source of bleeding was identified, post-operative CT abdomen was carried out which showed minimal fluid with heterogeneous enhancement of the uterus. 23x15 mm enhancing soft tissue structure in left adnexa this looked edematous.

Diagnosis of Interstitial Pregnancy was suspected.

Patient remained in ICU and given supportive treatment, β hcg done which increased to 919 IU therefore Methotrexate IM was given. Patient developed pancytopenia after methotrexate injection. She was managed by multidisciplinary team of haematologist, intensivists and physician and discharged home in stable condition on 12th post op day.

Case 5

38-year-old, Moroccan lady, G4P2+1, 7 weeks pregnant, referred from Health Centre on 14/8/2023, with abdominal pain, colicky in nature. She was seen and evaluated by medical officer in emergency department. Patient was vitally stable. TVS done in emergency showed bilateral adnexal mass and intrauterine gestational sac of 5.4 weeks, with no fetal pole. Routine laboratory examination and β hcg were sent and advised patient to wait for the results. She could not wait for the results and attended next day on 15/8/2023, when she was having diffuse abdominal pain but was vitally stable. Beta hcg which was 35459miu/ml. Specialist was called to review the case. TVS was done which showed thickened endometrium, with left adnexal mass with intrauterine gestation sac seen with fetal pole. The right ovary looked normal with moderate amount of free fluid in pouch of Douglas. Impression was ruptured ectopic pregnancy and plan was made for immediate laparotomy. Laparotomy findings suggested moderate hemoperitoneum, bulky uterus, left ruptured ectopic, right tube with bluish bulge 1 inch from the fimbrial end, looked like ectopic pregnancy. Both ovaries were normal. Left salpingectomy was done, Right salpingostomy done and ectopic tissue was removed and sent for histopathology. Patient remained stable post operatively. β hcg repeated after 48 hours came as 3170 m IU/ml. Patient was given methotrexate and weekly follow up appointment. Diagnosis was confirmed as spontaneous bilateral ectopic pregnancy by histopathology.

Discussion:

Unusual sites account for approximately 3.2% of all ectopic in our hospital.

Primary ovarian pregnancy is one of the rarest forms of ectopic pregnancy. The exact cause remains unknown. However, IVF, IUCD and other factors such as endometriosis, previous adnexal surgeries, CS, previous infectious diseases, history of infertility has been implicated.⁽³⁻⁶⁾

Previous two CS might be the cause in our patient.

The signs and symptoms of ovarian pregnancy are similar to ruptured tubal pregnancy, hemorrhagic corpus luteum cyst, or chocolate cyst.

There is no specific criteria to make a preoperative diagnosis and remains a challenge and detection is often made at the time of surgery and confirmed by the histological examination.

Ultrasound scan findings of fetal sac inside the ovary surrounded by ovarian tissue is most suggestive findings⁽⁷⁾

Most preferred treatment is the removal of the products of conception or wedge resection of ovary.⁽⁸⁾

In our case we removed the product of conception completely and reconstructed the ovary.

However, there are some reported cases of use of systemic methotrexate if the risk of surgery to the patient is presumably high or in the presence of persistently raised beta hcg levels postoperatively.⁽⁹⁾

Caesarean scar pregnancy (CSP) increasing with the increasing incidence of caesarean section rate.

The gestational sac is fully or partially implanted within the niche of previous caesarean scar. The most probable mechanism is invasion by the implanting embryo over the previous scar caesarean.⁽¹⁰⁾

Approximately thirteen percent of cases are misdiagnosed as are confused with implantation in lower segment or miscarrying event or cervical pregnancies.⁽¹¹⁾

In our hospital in last 3 years we have managed 4 cases of scar site ectopic. One of our case was misdiagnosed initially who had three previous LSCS. Most likely the initial ultrasound was ignored or was unclear.

Ultrasound criteria for the diagnosis include.⁽¹²⁻¹⁶⁾

1. Empty uterine cavity
2. Gestational sac located at internal os embedded at the site of the previous CS scar.
3. Thin or absent layer of myometrium between the gestational sac and the bladder.
4. Evidence of prominent trophoblastic/Placental circulation on colour Doppler examination.
5. Empty endocervical canal.

MRI may be of use in some cases where diagnosis by ultrasound appears to be difficult.⁽¹⁷⁾

The most preferred mode of treatment for CSP has not been defined. There is a potential risk of hemorrhage and subsequent hysterectomy. In most of the available treatment considered for CSP. Therefore, treatment should be individualized after proper evaluation.

Expectant management has very high failure rate therefore usually not recommended. Medical management with methotrexate may be considered in hemodynamically stable women here fetal hear beat not present whereas surgical treatment should be considered for unstable women or who decline medical treatment.

In our case patient opted for medical treatment and had a successful outcome.

Incidence of **Heterotopic pregnancy** is increased from approximately 1 in 40,000 with pregnancy to 1 in 100 due to increasing use of assisted reproductive technologies (ART) widely. ⁽¹⁸⁻¹⁹⁾

In our case the diagnosis was misdiagnosed initially, as often when intrauterine pregnancy are established the thought of having heterotopic pregnancy is unlikely ⁽²⁰⁾

The management for heterotopic pregnancy depends on the wishes of the woman and general condition at the time of presentation. ⁽²¹⁾

In our case the patient opted for salpingectomy leaving the intrauterine pregnancy intact as methotrexate was not an option for her.

Clinicians must remain alert to the fact that confirming an intrauterine pregnancy does not exclude the coexistence of ectopic pregnancy where patients present with acute symptoms nor related to tubal pregnancy (intact with other signs of rupture or bleeding). Both tubes and ovaries should be routinely evaluated.

Interstitial pregnancy is a rare often presents with to uterine rupture resulting in life-threatening haemorrhage⁽²²⁾ Early diagnosis is crucial to reduce morbidity and mortality. Ultrasound findings may suggest empty uterine cavity, with a mass/gestational sac located laterally in the interstitial (intramural) part of the tube. Often the sac is surrounded by myometrium in all imaging plane. ⁽⁹⁾

USS findings were present in our case (Fig 10) but was missed. MRI can be helpful in the confirmation of the diagnosis if proper diagnosis is not possible as done in our case.

Surgical management by laparoscopic cornual resection or salpingotomy is an option in some cases. Alternative surgical techniques include hysteroscopic resection under laparoscopic or ultrasound guidance. ⁽⁹⁾ Our patient was offered systemic methotrexate after laparotomy.

Spontaneous Bilateral Ectopic pregnancy is the rarest form of extra uterine pregnancy. ^{(23) (24, 25)}

It is most likely associated with multiple ovulations, transperitoneal migration of trophoblastic tissue from one tube to the other and superfetation. ^(26,27)

The most probable mechanism in our case is adhesion due to previous 2 LSCS which hamper tubal motility leading to ectopic. The diagnosis is only confirmed by findings of product of conception from both tubes on histopathological examination. ^(25, 28)

There are no well-defined studies or data to suggest standard of care in the case of bilateral ectopic pregnancy especially when one tube is ruptured and other is unruptured. In our case we offered surgical and medical both, because of her future fertility requirement. Salpingectomy for the ruptured tube and salpingostomy for the unruptured tube and gave her methotrexate post operatively for any residual trophoblastic tissue, in order to give her a maximum chance to preserve fertility.

Conclusion: Rare and atypical cases of ectopic pregnancies pose significant diagnostic and therapeutic challenge. Early diagnosis may help to choose the proper management. A high index of suspicion and early and timely involvement of senior assistance is the key to decrease the related maternal morbidity and mortality.

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Fig 1: Showing Right ovarian corpus luteal Cyst & left ovarian ectopic pregnancy



Figure 2 Specimen from Rt. ovarian cyst and left ovarian ectopic



Figure 3: TAS Scar Site ectopic, GS embedded ectopic mass



Figure: 4 Right fallopian tube totally distorted by ectopic



Figure:5 3D USG showing viable intrauterine pregnancy



Figure6: Empty endometrium, IUCD in situ, small ectopic at the left cornual