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Ovarian Ectopic Pregnancy: A Rare Case Report in a Resource-Limited Setting.

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ABSTRACT

Background: Ovarian ectopic pregnancy (OEP) is a rare form of ectopic pregnancy which occurs when a fertilized ovum implants into the ovary. The most common risk factor is prior use of IUCD. Management of OEP can be conservative or surgical.

Case: We report a 32-year-old Gravida 2 Para 0010 with a history of right ectopic pregnancy successfully managed medically two years ago who came presenting with severe right lower abdominal pain and 08 weeks amenorrhea. Pregnancy test was positive, laparotomy revealed presence of an unruptured right fluid filled ovarian mass which was removed through wedge resection.

Conclusion: Ovarian ectopic pregnancy should be considered in all reproductive age women who present with severe acute abdominal pain and positive pregnancy test even if ultrasound findings are inconclusive especially in low resource setting were quantitative serum β HCG and laparoscopy are not available.

Keywords: Ovarian ectopic pregnancy, laparotomy, case report, low resource setting.

INTRODUCTION

Ectopic pregnancy is a form of pregnancy in which the embryo attaches itself outside of the uterus, most commonly (95% of cases) in the fallopian tubes [1]. Ovarian ectopic pregnancy (OEP) is a rare form of ectopic pregnancy which occurs when a fertilized ovum implants into the ovary [2]. It occurs in 0.5% to 3% of most ectopic cases. It's incidence ranges between 1 in 7,000 to 1 in 40,000 live births [3].

The most significant risk factor for OEP are intrauterine contraceptive devices (IUCD) which accounts for approximately 57% to 90% of patients with primary OEP [4,5]. Other risk factors include pelvic inflammatory disease (PID), sexually transmitted infections (STIs), endometriosis, previous ectopic pregnancy, use of assisted reproductive technologies, prior pelvic surgery, advanced maternal age, multiparity [2,6].

Diagnostic evaluation in OEP include serial quantification of serum beta HCG level measurement [7], Transvaginal ultrasound (TV-US) which reveals a wide echogenic ring with an internal echo lucent area on the ovarian surface ("ring of fire" sign) [6], laparoscopy remains the gold standard for the diagnosis of OEP [8]. MRI has also been used in some reported cases of OEP for localizing the implantation site when it is unclear on TV-US [9].

The management of OEP can be medical or surgical. Medical management can be done using a single dose of intramuscular methotrexate, though uncommon and controversial, there is limited evidence available regarding management of OEP with methotrexate even though some cases have reported successful results [4]. Surgical management with laparoscopy remains the gold standard due to its minimally invasive nature where a wedge resection, cystectomy or oophorectomy is performed depending on age, patients desire to conceive and others [8]. Laparatomy is done in hemodynamically instable patients. Histopathologic examination can be performed post operatively to confirm the diagnosis [8].

In low resource setting with third category hospitals, the diagnosis and management of OEP remains very challenging due to lack of adequate paraclinical such as quantitative beta HCG serum levels and laparoscopy. Surgical management in low resource setting is mostly done through laparotomy which has the inconvenience of being highly invasive and increases the risk of postoperative complications.

CASE PRESENTATION

This is a 32-year Gravida 2 Para 0010, with a contributive history of right ectopic pregnancy two years ago which was managed medically and last menstrual period (LMP) of 08 weeks prior to consultation who came presenting with severe right lower abdominal pain, burning in nature, continuous, non-radiating with no relieving or aggravating factor. There was no per vaginal bleeding, no vaginal discharges. Review of system was remarkable for one episode of vomiting. On physical examination the patient was seen in pains holding the right iliac fossa with BP: 105/66mmHg, pulse: 87 BPM, Temp0: 36.80C, the conjunctiva was pink, abdomen non distended, tenderness at the hypogastric and right iliac fossa, no rebound tenderness, tympanic abdomen with bowel sounds active.

On vaginal examination, the vulva was clean and the cervix was closed. Our initial diagnosis was a right ectopic pregnancy based on the severe right sided pain and LMP of 08 weeks. Our differential diagnosis was a ruptured right hemorrhagic cyst and appendicitis. Laboratory investigation revealed a positive urine pregnancy test (Qualitative βHCG), quantitative β HCG was unavailable in our setting, full blood count was normal. For the radiologic investigation, a pelvic ultrasound was done which revealed an empty uterus (Figure 1), an unruptured tubal (ampullary) ectopic pregnancy of 09 weeks 03 days (Figure 2), free fluid in the pouch of Douglas and an unruptured right ovarian cyst of approximately 30 ml (Figure 3). A transvaginal ultrasound and laparoscopy were not available in our setting.

Due to the severe right lower abdominal pain the patient experienced, associated with the diagnostic uncertainty between the unruptured ectopic pregnancy and the unruptured ovarian cyst, we opted for a surgical management in first intention. An emergency laparotomy was done under general anesthesia, intraoperative findings revealed a non-gravid uterus, fallopian tubes were free with normal size, presence of a right fluid filled ovarian mass (figure 3, 4, 5) which was resected. Upon sectioning of the ovarian cyst, an encapsulated non-living fetus of approximately 09 weeks was seen with a surrounding clear amniotic fluid (figure 6, 7, 8).

In post operative management, the patient was hospitalized for a total of 05 days where she received intravenous antibiotics (ceftriaxone and metronidazole), analgesics (ketoprofen and paracetamol) with no post operative complications. One month after discharge from the hospital, the patient was asked to do STI related laboratory investigations to rule out pelvic inflammatory disease as one of the etiologies of the OEP and the treatment was done accordingly (see table 1). The patient was later referred to a gynecologist in town for better follow up and preparation for the next pregnancy.

DISCUSSION

Ovarian ectopic pregnancy (OEP) is a rare form of ectopic pregnancy which occurs when a fertilized ovum implants into the ovary [2]. It occurs in 0.5% to 3% of most ectopic cases. It's incidence ranges between 1 in 7,000 to 1 in 40,000 live births [3]. The first documented case of ovarian pregnancy was described in 1682 [10]. This is a case of a 32-year-old Gravida 2 Para 0010 who was successfully managed for ovarian ectopic pregnancy.

The most established and well recognized risk factor for OEP is prior use of the intrauterine device which ranges from 57% to 90% of patients with primary OEP [4,5]. IUD impairs tubal mobility, hence facilitating ovarian implantation. Other causes include pelvic inflammatory disease (PID), sexually transmitted infections (STIs), endometriosis, prior pelvic surgery and others. In our case, our patient had a history of ectopic pregnancy which was successfully managed medically [2,6]. In recent years, invitro fertilization has become a rising risk factor for OEP which can be explained by the application of ovulation stimulation drugs which increase sex hormone by the ovaries, hence affecting the contractile sensitivity of the uterine smooth muscle and interfering with the functioning of the uterus. After embryo transfer, the zygote moves back into the fallopian tube and implants itself in the ovary [11–13]. endometriosis and pelvic adhesions can block ovulation, forcing the egg cell to stay in the ruptured follicle and fertilization occurring directly in the ovary [14].

Although the exact pathophysiology of OEP is unknown, reflux of the fertilized egg back into the ovary is the most frequently known mechanism. Other methods that have been suggested including interfering with the release of the ovum from the broken follicle, fallopian tube dysfunction and inflammatory thickening of the ovarian tunica albuginea. Pathogenesis may also include fertilization out of the fallopian tube then implantation inside the ovarian stoma. The tunica albuginea is a structure that covers the ovary, lacking muscle fibers and filled with blood vessels and loose connective tissue. The tendency for early rupture may be exacerbated by this lack of muscle support. When the trophoblastic tissue invades the ovarian stroma, it damages nearby blood vessels, which causes intra-abdominal blood to quickly build up and rupture. This explains why hemoperitoneum and, occasionally hemodynamic instability are commonly found in ovarian pregnancies. Furthermore, the ovary's lack of decidualized endometrium may restrict the tissue's ability to support implantation, which would further increase the risk of early rupture [15].

The clinical manifestations of OEP are very similar to that of tubal ectopic pregnancy which include amenorrhea, abdominal pain and per vaginal bleeding. This shared symptomatology between OEP and tubal ectopic pregnancy makes it extremely hard to differentiate them preoperatively [16]. In our case, the patient had amenorrhea and pelvic pain only. When the ovarian pregnancy ruptures, the patient experiences severe abdominal pain due to large intra-abdominal hemorrhage begins which can be manifested clinically with hemorrhagic shock and anemia. The differential diagnosis of

OEP include tubal ectopic pregnancy, hemorrhagic ovarian cyst, ovarian torsion, tubo-ovarian abscess, acute appendicitis [2].

An elevated serum β HCG indicates the patient is pregnant even in other forms of ectopic pregnancy. Serum HCG continues to rise if treatment is started early enough but the value rarely doubles above 48 hours [20]. In low resource setting, quantitative serum HCG is hardly available as in our case were we mainly relied on qualitative β HCG, making follow up and therapeutic decisions very complicated. Laparoscopy remains a very good alternative for the diagnosis of OEP [21] however because most OEP are emergency consultations due to severe abdominal pain and hemodynamic instability, transvaginal ultrasound has become the main diagnostic modality because of its high availability and affordability especially in low resource settings. In our case a pelvic ultrasound was done due to the unavailability of the transvaginal probe. Recommended ultrasound finding for the diagnosis of ovarian pregnancy include 1) the presence of a wide echogenic ring with an internal echo lucent area on the ovarian surface, 2) the presence of an ovarian cortex, including a corpus luteum or follicles around the mass, and 3) the echogenicity of the ring being greater than that of the ovary itself [22–24].

Other imaging modalities include MRI which is mostly used when transvaginal ultrasound findings are insufficient. It visualizes the presence of extra uterine gestation structures that typically appear as a high-intensity mass containing distinct, low-intensity foci on T2-weighted imaging, which indicate hemorrhage [25]. CT scan is not routinely used in diagnosing OEP however it is important in the diagnosis of ectopic pregnancy at specific sites such as retroperitoneal [26], hepatic [27] and omental pregnancies [28]. Its use in diagnosing ovarian pregnancy remains limited.

The management of ovarian ectopic pregnancy can be conservative or surgical. Conservative treatment includes systemic methotrexate using either single dose [29] or multiple dose [30] regimens . Successful treatment of OEP with laparoscopic or intravaginal injection of methotrexate directly in the ovarian ectopic pregnancy has already been reported [31,32]. Conservative management is mainly done in patients who are hemodynamically stable and requires close monitoring and serial quantification of serum β HCG level to evaluate treatment progression. In our case, conservative management could not be done because we were able to quantify serum β HCG level for close monitoring and the patient experienced severe lower abdominal pain. Surgical management is done mainly through laparoscopy as gold standard because of its minimally invasive nature and less complications, but this requires the patient to be hemodynamically stable [33].

The German gynaecologist, Spiegelberg in 1878 proposed a criterion for the diagnosis of ovarian ectopic pregnancy. 1) The ipsilateral tube must be intact, 2) the gestational sac must occupy a position in the ovary, 3) the ovary must be attached to the uterus through the utero-ovarian ligament, and 4) there must be ovarian tissue attached to the pregnancy in the specimen [17]. OEP can be classified as intrafollicular or extrafollicular OEP based on the etiology [18]; In the intrafollicular OEP, the oocyte is not discharged from the follicle during ovulation. Subsequently, the sperm enters through the ruptured opening and initiates fertilization in the follicle [19]. Extrafollicular OEP occurs when an oocyte has been released from the follicle but becomes implanted on the ovarian surface after fertilization.

It mostly involves an ovarian wedge resection to remove as little normal ovarian tissue as possible [34]. In patients with hemodynamic instability due to massive abdominal bleeding an emergency laparotomy is a suitable option. In our case, conservative management could not be done because we were unable to quantify serum β HCG level for close monitoring and the patient experienced severe lower abdominal pain. Laparoscopy could not be done due to the lack of the laparoscopic machine as well as technical expertise to use it, we therefore went for a laparotomy where we did an ovarian wedge resection leaving the ovaries intact.

CONCLUSION

Ovarian ectopic pregnancy is rare but poses significant diagnostic and management challenges due to similar clinical symptomatology with tubal ectopic pregnancy and other differential diagnosis such as ovarian cyst, ovarian torsion and appendicitis. It is potentially life threatening when not diagnosed early and clinical recognition is necessary to reduce mortality and morbidity. Ovarian ectopic pregnancy should be considered in all reproductive age women who present with severe acute abdominal pain and positive pregnancy test even if ultrasound findings are inconclusive especially in low resource setting were quantitative serum β HCG and laparoscopy are not available.

PATIENT'S CONSENT STATEMENT

A clear and well drafted patient consent form has been attached on a separate word document highlighting the patient's signature and agreement to this case report.

AUTHOR CONTRIBUTIONS (CRediT)

Concept and Design: Sankara Nykam, Tchinda Dimitri, Fotio Theophile
Acquisition and Interpretation of Data: Steve Kouam, Nges Samuel
Drafting of Manuscript: Sankara Nykam, Tchinda Dimitri
Manuscript Review : Tatapong Lily Funzeh , Ma-Fese Dorcas Akwo
Supervision: Fotio Theophile
All the authors approve the final version of the manuscript.

CONFLICT OF INTEREST

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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TABLES

Table 1: STI related exams with their results.

STI exams	Results
Chlamydia serology	IgM: Negative, IgG: Negative
TPHA/VDRL	Negative
Herpes I and II	Negative
Vaginal smear culture	Bacteria isolated: Escherichia coli Sensitivity: ofloxacin, doxycycline, ciprofloxacin
Mycoplasma culture	Sterile

Figures

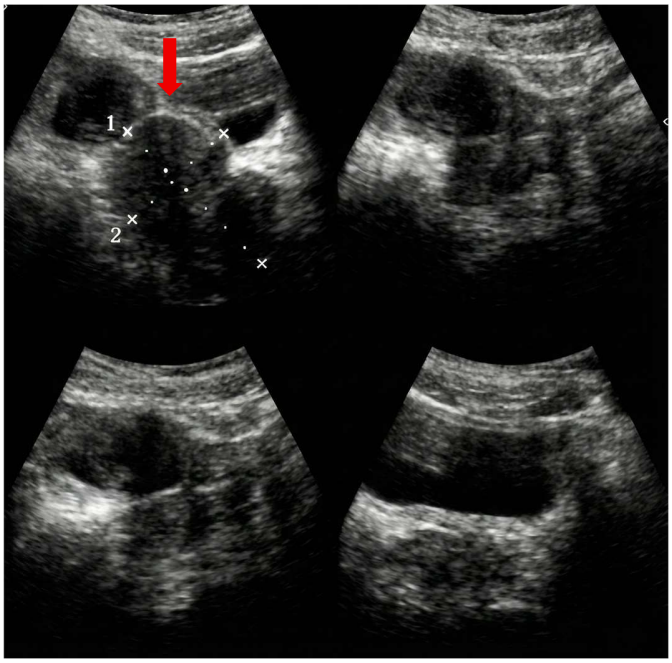

Figure 1: Pelvic ultrasound showing an empty uterus (red arrow).

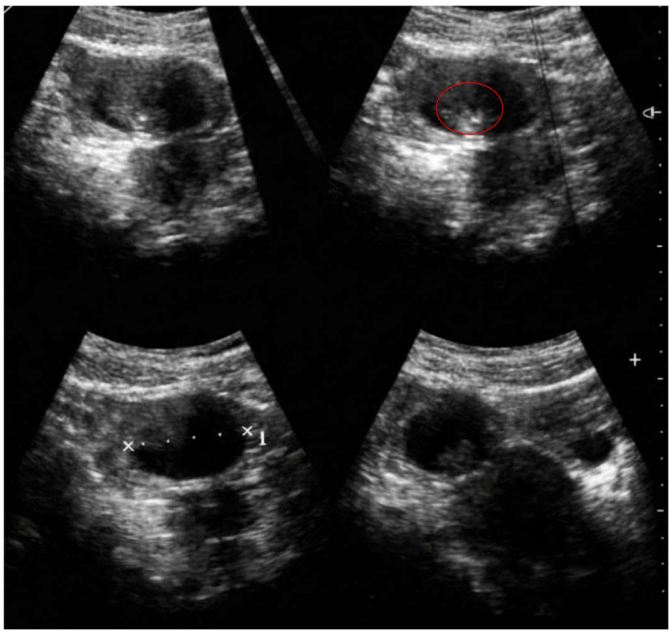

Figure 2: Pelvic ultrasound showing an annexial ectopic pregnancy of 09 weeks 03 days (red circle).


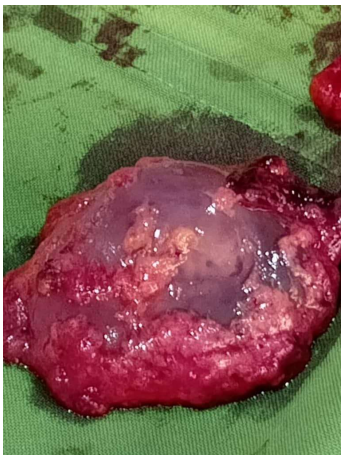


Figure 3, 4, 5: presence of a right fluid filled ovarian mass which was removed, with the ovary still intact.


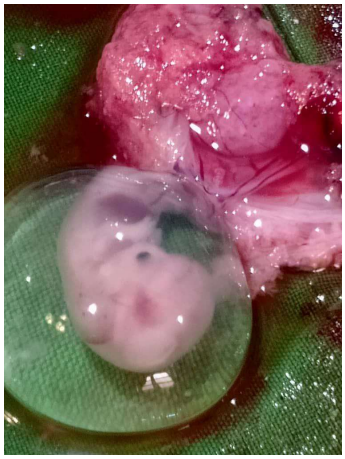
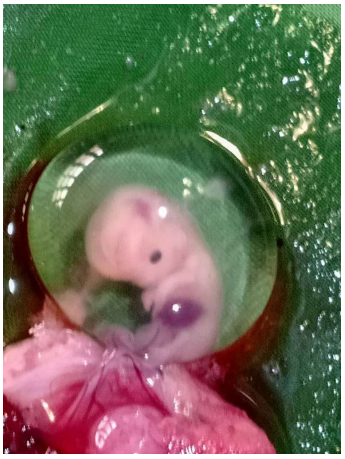

Figure 6, 7, 8: An encapsulated non-living fetus of approximately 09 weeks with a surrounding clear amniotic fluid.

Disclaimer

The ultrasound images used in this case report were color graded and enhanced using Google's Nano banana pro for removal of artifacts and high format resolution PNG format. The original echography file is shown below as proof of originality.