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Luteoma in ovarian ectopic pregnancy

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Abstract
Background: Pregnancy luteoma is a rare non neoplastic condition of the ovary. It is usually asymptomatic and found incidentally during imaging in pregnancy or during cesarean section. Pregnancy luteoma can also occur alongside ectopic pregnancy.
Case Presentation: We report the case of a 33 year old female presented with 6 weeks of ectopic gestation. The asymptomatic patient on routine USG investigations revealed right ovarian ectopic gestation. Laproscopic excision of the ovarian pregnancy was done. Histopathological examination confirmed ovarian gestation and in addition, the ovarian mass revealed solid aggregates of large cells with abundant eosinophilic cytoplasm; diagnosis of ovarian pregnancy with luteoma was given.
Conclusion: Ovarian pregnancy is rare; although awareness of this condition is important for reducing its associated morbidity and mortality. Also, luteoma must be considered in the differential diagnosis of ovarian masses in ovarian ectopic pregnancies that diagnosis of this entity may avoid unnecessary radical surgery.

Keywords: Ovarian pregnancy, ovarian masses, pregnancy luteoma

Introduction
Primary ovarian pregnancy (OP), where the gestational sac is implanted in the ovary, is one of the rarest forms of ectopic pregnancy. Its incidence after natural conception ranges from 1/2000 to 1/60 000 deliveries, accounting for 3% of all ectopic pregnancies [1]. Because of the increased vascularity of ovarian tissue, OP usually results in rupture and haemoperitoneum, making it a life-threatening gynaecological emergency. Use of transvaginal sonography has resulted in a more frequent diagnosis of unruptured ovarian pregnancies [2-3].
Pregnancy luteoma is a rare ovarian mass that develops during pregnancy and regresses after delivery [4]. It is a rare tumor that has received only scant mention in the general obstetric literature. The imaging features of this entity have been even less frequently reported. Generally, these masses are discovered incidentally during cesarean delivery or tubal ligation [5]. Given the rarity of luteoma, patients have unfortunately undergone cystectomies or oophorectomies by surgeons unfamiliar with the condition. Pregnancy luteomas depend on hCG stimulation during pregnancy for their structural and functional integrity. These mimic other ovarian masses predominantly the solid and complex cystic masses [6]. Good clinical and imaging features of pregnancy luteoma can obviate the requirement of an unnecessary surgery or termination of pregnancy. Though a rare event, pregnancy luteomas are reported along with ectopic pregnancies also [7]. To the best of our knowledge, the present study is the first to report luteoma in ovarian pregnancy. We describe the case of a 33-year-old woman with luteoma of ovarian pregnancy that was incidentally discovered during routine obstetric ultrasound.

Case Presentation
A 33-year-old woman, G2P1 was presented for routine obstetric ultrasound at 6 weeks of gestation. The right adnexa were tender to palpation though the patient had no signs of abdominal pain or pelvic bleeding. Beta hCG revealed plateauing results. She did not have any past history of pelvic inflammatory disease or insertion of an intrauterine device. The patient was hemodynamically stable and transvaginal sonography was done for the first initial impression of an ectopic pregnancy. TVS showed a uterus of normal size with no gestational sac but an echogenic mass medially to the right ovary of 9×11 mm size was identified which was suggestive of right ovarian ectopic pregnancy. Sonographically, another hypoechoic predominantly solid right ovarian mass containing multiple small cystic areas was identified.

It arose from the right ovary and compressed ovarian tissue to the periphery. The left adnexa were normal and there was no sign of free fluid in the abdominal cavity. Emergency laparoscopic excision of the ovarian pregnancy was performed. The gestational sac was easily shelled out of the encasing ovarian tissue. Pathologic analysis confirmed ovarian gestation with presence of necrotic chorionic villi and a corpus luteum within the ovarian tissue. The patient had an uneventful post-operative course and was discharged two days later.

Pathological findings

Histopathological examination showed chorionic villi, corpus luteum and ovarian stroma. Diagnosis of right sided ovarian pregnancy was confirmed. Microscopic examination also revealed inclusion cyst, corpus luteal cyst and focal aggregates of luteinized cells. Ovarian sections also showed sharply circumscribed mass of cells arranged in solid growth pattern replacing the normal ovarian parenchyma (Fig 1). The cells were moderate in size with abundant eosinophilic cytoplasm and central nuclei (Fig 2). Few of them showed nucleoli. Features were those of pregnancy luteoma.

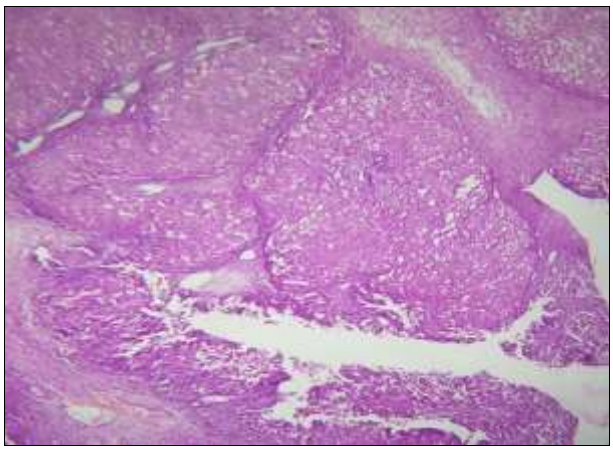

Fig 1: Well circumscribed solid mass of luteinized cells replacing ovarian tissue

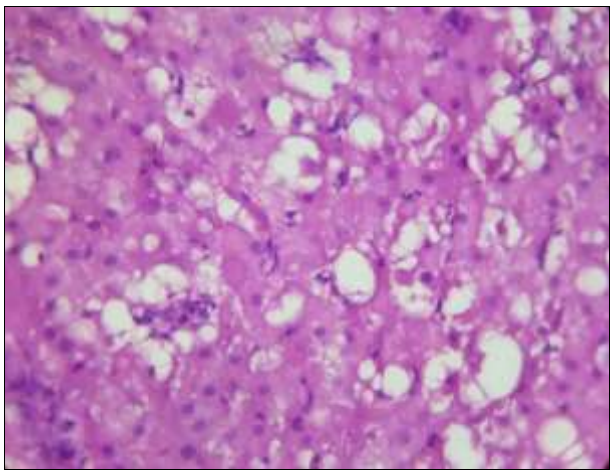

Fig 2: The cells moderate in size with abundant eosinophilic cytoplasm and central nuclei.

Discussion

Ovarian pregnancy is a rare variant of ectopic pregnancy. Early diagnosis of ovarian pregnancy is necessary in order to avoid more serious complications and emergency invasive procedures. An accurate differential diagnosis is important in ectopic pregnancies as patient management often differs depending on the type and exact location of the pregnancy [8].

Increased OP risk may be associated with factors such as endometriosis, previous adnexal surgeries, previous infectious diseases; history of infertility, in vitro fertilisation and embryo transfer (IVF-ET), polycystic ovarian syndrome and intrauterine device (IUD) use [9]. Whether these factors play etiological roles in the increasing occurrence of OP remains debated. However our patient did not have past history of any of the conditions discussed above and we hold up to the debate that the exact risk factors for OP remain to be ascertained.

In the past, OP has been treated by laparotomy and ovariotomy. However, conservative surgery, such as cystectomy or wedge resection, has proven to be a much safer and beneficial alternative [10]. Due to technical advances in laparoscopic surgery, laparoscopic treatment of OP with wedge resection can be viewed as the treatment of choice in select and hemodynamically stable patients and same has been performed in our patient [11].

Spigelberg criteria are historically used for intraoperative diagnosis of OP: intact fallopian tube on the affected side, fetal sac must occupy the position of the ovary on the affected side, ovary connected to the uterus by ovarian ligament; ovarian tissue must be located in the sac wall, which is confirmed by histopathology [12].

Luteoma associated with ectopic pregnancy is a rare condition and can mimic a solid ovarian neoplasm [7]. Incidental pregnancy luteoma has been reported in ovary submitted for ruptured ectopic tubal pregnancy. It was non encapsulated proliferation of thecal cells in the wall of atretic follicle and had all the pathological findings of late pregnancy luteoma. Many cases of pregnancy luteoma have been reported [13]. It is previously reported that pregnancy luteomas rarely present during early pregnancy [4]. However our patient with unruptured ectopic pregnancy was on her 6th week of gestation.

Luteoma of pregnancy must be distinguished from other ovarian masses to avoid oophorectomy in pregnant females. Differential diagnosis includes granulosa cell tumor, thecomas, stromal hyperthecosis, stromal luteomas and hyperreactio luteinalis [7]. Close clinical monitoring and appropriate follow up is recommended in case of strong clinical suspicion of pregnancy luteoma to avoid any radical surgery.

Conclusion

Ovarian pregnancy is a rare condition and its diagnosis is difficult and relies on criteria based on intraoperative findings. Its management remains surgical therapy; often conservative in hemodynamically stable patients with unruptured EP. Now, with ultrasonographic advances, OP can be diagnosed early, leading to conservative treatment and preservative surgery.

Ovarian pregnancy is acknowledged a rare event and luteoma associated with it is even rarest. Awareness of pregnancy luteomas is of paramount importance for keeping a higher index suspicion and considering them in the list of differential diagnosis for tubo ovarian masses. Antenatal accurate diagnosis is challenging but extremely important in order to optimize the obstetrical management of the patient. As pregnancy luteomas regress spontaneously, a more vigilant clinical outlook may help avoid unnecessary surgery, preserve fertility wherever possible and reduce patient morbidity.

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