Case Report

Retroperitoneal ectopic pregnancy after in vitro fertilization: A case report of a patient with bilateral salpingectomy

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ABSTRACT

Retroperitoneal ectopic pregnancy (REP) is a rare obstetric condition caused by the mislocalization of the gestational mass. The unexpected location often results in missed or delayed diagnoses, which can complicate the treatment process. We report the case of a 34-year-old Asian woman who presented to the hospital 31 days after embryo transfer with mild vaginal bleeding. A history of bilateral salpingectomies was established. Two operations were performed before we were able to successfully remove the gestational sac from the retroperitoneal cavity. The histologic finding suggested an interesting migration pathway for the pregnancy. REP should be considered when a visible gestational sac cannot be detected on ultrasound in the expected locations, particularly among patients who undergo treatment using assisted reproductive techniques (ART), and have a history of bilateral salpingectomies. Magnetic resonance imaging (MRI) plays a vital role in diagnosing REP and guiding surgical interventions. A multidisciplinary team is necessary to treat REP, and monitoring beta-human chorionic gonadotropin (βHCG) levels and histologic findings remain essential during follow-up.

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Background

Ectopic pregnancy is a complication encountered during the early stages of pregnancy, accounting for approximately 2% of all pregnancies [1]. Diagnosis is confirmed when the gestational sac is identified as implanting outside of the uterine cavity. The most common location for ectopic pregnancies is the Fallopian tubes, whereas abdominal implantation is much rarer, accounting for merely 1% [2], and retroperitoneal ectopic pregnancies (REPs) are even less common. Although an increasing number of REP cases have recently been reported, fewer than 30 cases have been reported in the English-language literature. Bizarre implantation locations can make diagnosis and treatment challenging, sometimes resulting in misdiagnosis, delayed treatment, and the application of passive management approaches, which can lead to catastrophic outcomes that threaten the patients’ lives [3]. The case presented in this report describes an REP that was initially misdiagnosed.

Case presentation

A previously healthy, 34-year-old Asian woman with gravida 3, para 2, presented at the hospital with vaginal bleeding. The patient reported a history of bilateral salpingectomies due to 2 previous tubal pregnancies, which resulted in the use of in vitro fertilization (IVF). Two 3-day-old embryos were transferred to the patient 31 days prior to hospital admission. Her vital signs were within normal limits, and she did not describe abdominal pain. A physical examination revealed no abdominal tenderness and no peritoneal signs. The bleeding status was mild, consistent with the patient’s stable overall performance. A quantitative beta-human chorionic gonadotropin (βHCG) test was indicated due to suspected ectopic pregnancy. The blood test revealed a highly elevated βHCG level of 19,182 UI/L. However, an emergency transvaginal ultrasound failed to identify any evidence of an intrauterine gestational sac, and the endometrial thickness was 7.5 mm. No adnexal mass was located. The patient was transferred to the Gynecologic Department (GD) for further follow-up and consultation due to her stable condition.

She was monitored for 2 days, which revealed a continued increase in βHCG levels, reaching 29,242 UI/L on day 3. A second comprehensive general abdominal ultrasound was performed, resulting in the discovery of a gestational sac near the right internal iliac artery. The gestational sac was approximately 20 × 25 mm and featured a visible yolk sac. The diagnosis of a presumed abdominal pregnancy indicated the patient for an emergent laparoscopy.

This first surgery revealed a normal uterus and intact ovaries. The Fallopian tubes had previously been completely removed, without evidence of fistulous leakage from the remaining stump. No intra-abdominal bleeding was detected, and the retroperitoneum remained intact. During the surgical procedure, the surgeons identified a small mass in the posterior abdominal wall, which was removed. Despite a lack of other indicators for pregnancy, we opted to evacuate the uterus. After surgery, the patient became stable, and was transferred back to GD for monitoring. Interestingly, the βHCG level, which was expected to decrease rapidly during the postoperative follow-up period, continued to increase gradually, reaching 36,386 UI/L on day 6, when the histologic findings were available. The pathologists did not identify any signs of trophoblast cells. The provided samples contained only endometrial tissue, and the small mass was diagnosed as unrelated endometriosis (Fig. 1). We immediately arranged for a second operation to locate the pregnancy using both magnetic resonance imaging (MRI) and ultrasound, which identified a suspected gestational sac near the right common iliac artery (Fig. 2). The sac was more significant than previous imaging findings.

During the second surgery, we converted from laparoscopy to laparotomy. A multidisciplinary team was formed, including gynecologists, general surgeons, and anesthesiologists. The exploration of the retroperitoneal cavity revealed a 2-cm gestational sac in close proximity to the right common iliac...
ectopic pregnancy. [1] Retroperitoneal pregnancy. [1] (changes in invasion and decrease post-operatively, out of the retroperitoneal space rather than on the peritoneal surface are even rarer [4]. Abdominal ectopic pregnancies, in general, and REPs, in particular, are associated with high mortality rates due to late diagnoses and unconventional treatments, which can include surgery or chemotherapy. As the use of assisted reproductive techniques (ART) has increased, reported REPs have also increased, and many reported REPs are associated with ART procedures, including embryo transfer (ET) after bilateral salpingectomies [5].

How the embryonic sac travels to the retroperitoneal cavity remains an unanswered question, particularly after invasive procedures, such as ET or bilateral salpingectomy. Three mechanistic theories have been proposed: the peritoneal, fistula, and lymphatic pathways [3].

The first theory refers to the peritoneal pathway and may occur during the performance of ET. In this proposed mechanism, embryos are misplaced in the retroperitoneal space either during or after the procedure due to uterine perforation. Perforations in both the uterine and peritoneal walls are necessary to facilitate gestational migration. However, in this case, the peritoneal wall, and uterus were intact. Moreover, the ET procedure was performed under strict ultrasonographic guidance, and is not likely to explain migration during gestation.

In the second theory, a fistula forms when the removed Fallopian tubes become covered with broad ligaments, creating a possible fistulous connection between the uterine, and retroperitoneal cavity. In our patient, the stumps of both Fallopian tubes were visible, intact, and detached from the broad ligaments, excluding this explanation.

The final theory suggests that the embryo migrates to the retroperitoneal space through lymphatic channels, similar to the metastasis of endometrial cancer, which may explain the ability of the embryo to travel such a long distance from the uterine cavity. An interesting example was reported in 2002 by Dmowski, who identified a gestational sac in the subpancreatic region [6]. This possibility appeared to be the most relevant mechanism in our case for 2 primary reasons. First, the general ultrasound performed on day 3 identified a gestational sac near the right internal iliac artery. However, on day 6, both the ultrasound, and MRI findings showed that the sac had migrated upward to the right common iliac artery. Second, the final histologic result provided indisputable evidence of the trophoblastic invasion of lymphatic tissues. To our knowledge, this represents the first case to report such findings.

Whether this case was successfully managed is difficult to assess, although some lessons can be learned. First, gynecologists should consider REP as a possible diagnosis when no gestational sac can be identified on transvaginal ultrasound. Many recently reported cases of REP are associated with ET and bilateral salpingectomies; therefore, close follow-up of such patients is crucial. In this particular case, an iatrogenic misdiagnosis resulted in 2 separate operations to evacuate the pregnancy, although this is not uncommon due to the unfamiliar location of the pregnancy [7].

Second, the initial misdiagnosis emphasized the essential role of MRI as an objective means for diagnosing REPs, and other extraterine pregnancies. MRI should be used early, especially in patients with highly elevated βHCG levels, and no corresponding intrauterine findings. MRI can provide detailed

**Fig. 3** – The second histologic finding identified the trophoblastic invasion of lymphatic tissue (star indicates lymphatic tissue, arrow indicates trophoblast cells).

![βhCG Level](image)

**Fig. 4** – Changes in the patient’s beta-human chorionic gonadotropin (βHCG) levels.

The patient remained able to support a successful pregnancy. Through IVF, she became pregnant again 1 year after the event and gave birth to a beautiful, healthy son.

### Discussion

Retroperitoneal pregnancies are extremely rare. Although approximately 2% of all pregnancies are ectopic [1], the incidence of abdominal pregnancies is as low as 1 in every 5000 ectopic pregnancies, and cases in which the sacs implant in the iliac artery. Fortunately, the sac had not invaded the large vessel, and the gestational sac was successfully removed without causing arterial damage and with minimal blood loss. Post-operatively, the quantitative βHCG test revealed a rapid decrease in βHCG levels, and the patient was safely discharged on day 10. A later pathology report illustrated the invasion of trophoblast cells into the lymph node tissue (Fig. 3). Serum βHCG concentrations were strictly monitored in the outpatient setting until no detection occurred after 4 weeks (changes in the βHCG levels over time are shown in Fig. 4).

The patient remained able to support a successful pregnancy. Through IVF, she became pregnant again 1 year after the event and gave birth to a beautiful, healthy son.
and comprehensive information regarding the location of the pregnancy, which is vital information in cases such as ours, in which the gestational sac is thought to have followed a lymphatic pathway, moving to a new location over just a few days. Knowing the location of the gestational sac can better prepare surgeons for the surgery. Moreover, MRI, and CT can be used not only for diagnostic purposes but also to guide treatment [8].

Immediately upon confirmation of an REP diagnosis, a multidisciplinary team, including gynecologists, general surgeons, and anesthesiologists, should be formed to determine the best treatment option for the patient. For cases in which surgical intervention appears to be the best option, a laparotomy should be considered over an endoscopy to provide more space for the surgeons to effectively remove chorionic invasions, particularly those attached to large vessels.

Finally, after the surgery, if the complete removal of trophoblastic invasion cannot be successfully achieved. The patient's βHCG levels must be closely monitored until they become entirely negative. Chemical interventions might be necessary in cases with suspected persisting lesions or unremovable masses [8–10].

Conclusions

Although REPs are extremely rare, they should be considered as one of the last diagnoses in patients with unexplainable increases in βHCG levels without a visible gestational sac on ultrasonography. This consideration is particularly important among those who have recently received ET and bilateral salpingectomies. Histology in these cases plays a crucial role in confirming the diagnosis. Combined sonographic and MRI findings are essential for diagnosis, and quantitative βHCG monitoring is necessary for patient follow-up. A multidisciplinary team should be formed for consultation and intervention upon diagnosis.

Authors’ contributions

NDA was responsible for the conception and design of the work as well as data analysis and interpretation. NDA, NXH, NTTTH, NKT, and PTHT were responsible for data collection. PTHT and NMD drafted the article, which was critically revised by all authors; and all authors were responsible for the final approval of the version to be published.

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Availability of data and material

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Consent for publication

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