



Successful Management of a Heterotopic Pregnancy Following Bilateral Salpingo-oophorectomy: A Case Report

Rania E Belal^{1*}, Jullia M Nageeb², Acha Assmanni Adam³

1. Specialist of obstetrics and Gynecology in Saudia Arabia, MD Obstetrics and Gynecology, Medical Council of Sudan, Membership of Royal College of Obstetricians & Gynaecologists-UK
2. Membership of Royal College of Physicians of Ireland Obstetrics & Gynaecology, Membership of Royal College of Obstetricians & Gynaecologists-UK
3. Consultant of obstetrics and Gynecology in Saudia Arabia, MD Obstetrics and Gynecology. Medical Council of Sudan, Membership of Royal College of Obstetricians & Gynaecologists-UK

***Correspondence to:** Rania E Belal, Specialist of obstetrics and Gynecology in Saudia Arabia, MD Obstetrics and Gynecology, Medical Council of Sudan, Membership of Royal College of Obstetricians & Gynaecologists-UK.

Copyright

© 2024 **Rania E Belal**. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Received: 16 October 2024

Published: 23 October 2024

DOI: <https://doi.org/10.5281/zenodo.13981111>

Abstract

Heterotopic pregnancy, characterized by the simultaneous presence of intrauterine and ectopic pregnancies, is a rare but significant complication in reproductive medicine, particularly among patients undergoing assisted reproductive technologies (ART). This case report documents the successful management of a heterotopic pregnancy following bilateral salpingo-oophorectomy in a 36-year-old woman. The patient, gravida 4, para 2 + 1, presented with mild vaginal bleeding and lower abdominal pain at 9 weeks gestation. Ultrasound examination revealed two gestational sacs, one intrauterine and one ectopic. Following laparoscopic surgery to address the ectopic pregnancy, the intrauterine pregnancy progressed normally and culminated in a successful vaginal delivery at term. This case showed the importance of early detection and comprehensive management in achieving positive outcomes for heterotopic pregnancies, even in patients with a history of significant reproductive surgical interventions. Further research is needed to refine diagnostic and treatment protocols for such high-risk cases.

Keywords: *Heterotopic pregnancy, bilateral salpingo-oophorectomy, ectopic pregnancy, intrauterine pregnancy, assisted reproductive technologies, early detection, ultrasound, laparoscopic surgery, case report.*

Introduction

Heterotopic pregnancy, characterized by the simultaneous presence of an intrauterine and an ectopic pregnancy, remains a rare but significant complication in reproductive medicine (1). This condition is particularly notable among patients undergoing assisted reproductive technologies (ART), such as in vitro fertilization (IVF). The incidence of heterotopic pregnancy in IVF pregnancies has been reported to be as high as 1% (2).

In a notable case study by Shavit et al. (3), a 35-year-old woman undergoing IVF treatment experienced two consecutive heterotopic pregnancies in the tubal stumps after a bilateral salpingectomy. Despite surgical removal and coagulation of the tubal stumps, the patient had recurrent heterotopic pregnancies, with both

intrauterine pregnancies ending in spontaneous abortions following surgical intervention for the ectopic pregnancies. Similarly, Barrenetxea et al. (4) documented cases of heterotopic pregnancies post-bilateral salpingectomy in IVF patients. Their findings emphasize that abdominal pain and vaginal bleeding in early pregnancy should raise suspicion for heterotopic pregnancy, even if an intrauterine pregnancy is confirmed, highlighting the importance of thorough ultrasound examinations in these scenarios.

Comparative studies have shown the increased risk and complexity associated with heterotopic pregnancies. Clayton et al. (5) found that intrauterine gestations in heterotopic pregnancies were more likely to end in spontaneous or induced abortions and were less likely to result in live births compared to intrauterine pregnancies alone. Dor et al. (2) further highlighted the necessity for vigilant monitoring and early diagnosis to manage such high-risk pregnancies effectively, given the incidence of combined intrauterine and ectopic pregnancies post-IVF and embryo transfer.

The complexity of managing heterotopic pregnancies is compounded by the presence of risk factors such as previous tubal damage, pelvic inflammatory disease, and prior ectopic pregnancies. Talbot et al. (6) found that 71% of women with heterotopic pregnancies had at least one risk factor for ectopic pregnancy, showing the need for personalized risk assessment and tailored management strategies in high-risk patients. Furthermore, Lim and Fuentes (7) reported a case of spontaneous intrauterine pregnancy post-bilateral salpingectomy, which highlights the potential for sterilization failure due to residual tubal tissue, particularly in the presence of pelvic adhesions. This finding showed the importance of thorough surgical techniques and postoperative monitoring to mitigate the risks of incomplete salpingectomy and subsequent pregnancies.

Recurrent heterotopic pregnancy following bilateral salpingo-oophorectomy, while exceedingly rare, remains a possibility (8 – 10). Surgical treatments for tubal disease, such as salpingectomy, do not entirely eliminate the risk of ectopic pregnancies due to potential residual tubal tissue capable of implantation. Shavit et al. (3) suggested that the presence of an intrauterine pregnancy often leads to a false sense of security, delaying the diagnosis of concurrent ectopic pregnancies. Therefore, heterotopic pregnancy should be considered in patients with abdominal pain or vaginal bleeding, regardless of a confirmed intrauterine pregnancy (12, 13). Management of heterotopic pregnancy typically involves surgical intervention to preserve the intrauterine pregnancy. However, success rates vary, with a reported 66-68% survival rate for intrauterine pregnancies in heterotopic cases as reported by Barrenetxea et al (4). Methotrexate is usually avoided to prevent compromising the viable intrauterine pregnancy, with potassium chloride or hyperosmolar injections as alternative treatments. The significant diagnostic and management challenges posed by heterotopic pregnancy following bilateral salpingo-oophorectomy necessitate comprehensive ultrasound examinations and high

clinical suspicion for early detection and appropriate management (14, 15). Future research should focus on refining diagnostic protocols and treatment strategies to improve outcomes for both intrauterine and ectopic components of heterotopic pregnancies.

The aim of this case report is to document the successful management of a heterotopic pregnancy following bilateral salpingo-oophorectomy, providing insights into diagnostic and therapeutic approaches. Given the rarity of such cases, this report adds valuable knowledge to the existing literature, emphasizing the importance of comprehensive diagnostic protocols and personalized management strategies.

This case report is expected to contribute to the medical literature by providing a detailed rare and complex clinical scenario, highlighting the diagnostic challenges and the need for high clinical suspicion in similar cases. It emphasized the importance of thorough postoperative monitoring and the potential for residual tubal tissue to cause ectopic pregnancies, offering insights into successful management strategies that could be employed in future cases. Moreover, the report aims to improve clinical practices and outcomes for patients facing similar medical conditions.

Case Presentation

A 36-year-old woman, gravida 4, para 2 + 1, presented to Al-Mowasat Hospital in Jubail Industrial City in the Eastern Province of the Kingdom of Saudi Arabia in 2020. Her obstetric history included two full-term normal vaginal deliveries, followed by the use of an intrauterine contraceptive device (IUCD) for five years. After the removal of the IUCD, she conceived spontaneously, which resulted in an ectopic pregnancy at 5 weeks gestation. This ectopic pregnancy was managed with right laparoscopic salpingo-oophorectomy following confirmation by ultrasound and MRI. The current case represents her fourth pregnancy, characterized by heterotopic pregnancy.

One year after her first ectopic pregnancy, the patient conceived spontaneously again. She experienced spotting early in the pregnancy, and an initial ultrasound at another hospital suggested the presence of a luteal cyst. At 9 weeks gestation, she presented to the emergency room with mild vaginal bleeding and lower abdominal pain. An ultrasound examination revealed two gestational sacs: one intrauterine and one extrauterine adnexal mass with a heterogeneous echopattern, raising suspicion for a tubal pregnancy. Based on these findings, she was admitted to the hospital and counseled regarding the need for surgical intervention. Further diagnostic workup for the current pregnancy included ultrasonography, which revealed the following: an anteverted and enlarged uterus displaying a single well-defined fundal gestational sac with a crown-rump length (CRL) of 24 mm, corresponding to approximately 9 weeks and 1 day of gestation, with an estimated

due date (EDD) of 2/03/2020. Fetal cardiac activity was confirmed by Doppler interrogation, showing a fetal heart rate (FHR) of 181 bpm. The gestational sac was surrounded by a normal decidual reaction, and the internal os was closed. Additionally, there was a left adnexal mass lesion with a heterogeneous echopattern measuring 37 mm x 33 mm, likely of tubal origin and suspected to be a left tubal ectopic pregnancy. Both ovaries appeared normal, and there was mild free fluid present in the cul-de-sac. Based on these findings, heterotopic pregnancy was highly considered.

Following the diagnosis of heterotopic pregnancy, the patient underwent laparoscopic surgery, which confirmed a left tubal ectopic pregnancy. Given the extensive damage to the tube, a left salpingo-oophorectomy was performed as the tube was unsuitable for salpingostomy. Postoperatively, she was hospitalized for two days for observation, which was uneventful. The patient was then discharged and continued to receive routine antenatal care. The intrauterine pregnancy progressed normally without complications until term.

At 40 weeks gestation, the patient presented with decreased fetal movement. Cardiotocography (CTG) indicated reduced fetal viability, leading to her admission for labor induction. She subsequently delivered vaginally without complications. Given that a past history of ectopic pregnancy is a risk factor for recurrence, the patient's only significant medical history was her previous ectopic pregnancies. She had no other notable medical or surgical history, and no additional risk factors for her current pregnancy complications were identified.

This case showed the importance of early detection and comprehensive management of heterotopic pregnancies, even in patients with a history of bilateral salpingectomy. Despite the challenges, the patient's intrauterine pregnancy was successfully carried to term, demonstrating the potential for positive outcomes with appropriate and timely treatment.

Discussion

Heterotopic pregnancy, particularly following bilateral salpingo-oophorectomy, is exceedingly rare. The incidence of heterotopic pregnancies in IVF patients can be as high as 1% as reported by Dor et al (2), but cases following bilateral salpingo-oophorectomy are even less common due to the nature of the surgery, which typically removes the primary sites for ectopic implantation. Risk factors include previous tubal damage, pelvic inflammatory disease, prior ectopic pregnancies, and the use of ART (6). This case aligns with the literature, where ART was a significant contributing factor to the development of heterotopic pregnancy despite the prior removal of the fallopian tubes.

The clinical presentation in this case involved abdominal pain and vaginal bleeding, symptoms that are consistent with those reported in similar cases (3, 4). These symptoms should always raise suspicion for a heterotopic pregnancy, especially in patients with known risk factors or those undergoing ART. Early ultrasound examinations are crucial in such scenarios to differentiate between intrauterine and ectopic components, ensuring timely diagnosis and intervention.

Complications associated with heterotopic pregnancies include spontaneous abortion of the intrauterine pregnancy, rupture of the ectopic site leading to hemorrhage, and potential loss of future fertility due to surgical interventions. In this case, the intrauterine pregnancy was successfully managed without spontaneous abortion, contrasting with the outcomes reported by Shavit et al. (3), where both intrauterine pregnancies ended in spontaneous abortions following surgical intervention for the ectopic pregnancies. The presence of residual tubal tissue post-surgery, as highlighted by Lim and Fuentes (7), remains a potential risk factor for recurrence.

Management of heterotopic pregnancy typically involves surgical intervention to remove the ectopic pregnancy while preserving the intrauterine pregnancy whenever possible. This case was managed successfully through surgical intervention, aligning with the strategies suggested in the literature (2, 4). Methotrexate was avoided to prevent compromising the viable intrauterine pregnancy, and alternative treatments such as potassium chloride or hyperosmolar injections were considered.

The success in preserving the intrauterine pregnancy in this case highlights the importance of tailored management approaches. Preventive measures include thorough surgical techniques during procedures like salpingo-oophorectomy to minimize the risk of residual tubal tissue, which can serve as a site for future ectopic pregnancies (Lim & Fuentes, 2024).

Counseling for patients undergoing ART should include discussions about the risk of heterotopic pregnancy and the importance of early monitoring. High clinical suspicion should be maintained in patients presenting with abdominal pain or vaginal bleeding, regardless of confirmed intrauterine pregnancy. This case contributed to the existing body of literature by providing an example of successful management of heterotopic pregnancy following bilateral salpingo-oophorectomy.

Unlike the case reported by Shavit et al. (3), where recurrent heterotopic pregnancies occurred, this case demonstrates that careful surgical intervention and vigilant monitoring can lead to positive outcomes. The comparative review by Barrenetxea et al. (4) and the findings by Clayton et al. (5) regarding the higher likelihood of spontaneous or induced abortions in heterotopic pregnancies showing the complexity of managing such cases. However, the successful preservation of the intrauterine pregnancy in this instance adds

a valuable perspective to the potential for favorable outcomes with appropriate management.

Thus, the successful management of this heterotopic pregnancy post-bilateral salpingo-oophorectomy showed the importance of early diagnosis, tailored surgical intervention, and thorough postoperative monitoring. This case showed the need for ongoing research to refine diagnostic and treatment protocols to improve outcomes for patients with similar high-risk profiles.

Conclusion

This case highlights the successful management of a heterotopic pregnancy in a patient with a history of bilateral salpingectomy. Despite the complexities associated with her condition, early detection and timely surgical intervention allowed for the preservation and successful delivery of the intrauterine pregnancy. The patient outcome showed the potential for positive results with diligent monitoring and comprehensive care in cases of heterotopic pregnancy, even in those with significant reproductive surgical histories.

Implications for Clinical Practice and Further Research

In clinical practice, this case emphasizes the importance of vigilant monitoring and early ultrasound evaluation in pregnant patients with a history of ectopic pregnancies and bilateral salpingectomy. Clinicians should maintain a high index of suspicion for heterotopic pregnancy, especially in patients presenting with abdominal pain or vaginal bleeding, regardless of the presence of an intrauterine gestation.

A comprehensive diagnostic workup, including detailed ultrasonography and possibly MRI, is crucial for accurate diagnosis and management. Identifying both intrauterine and ectopic components early can prevent complications and guide appropriate intervention. The individualized management plan in this case, which included laparoscopic salpingo-oophorectomy due to the extensive damage to the fallopian tube, showed the need for tailored treatment strategies based on the patient history and current presentation. Postoperative care and close follow-up are essential to ensure the well-being of both the mother and the fetus, necessitating regular antenatal visits and timely interventions if complications arise.

Further research is needed to better understand the incidence and risk factors associated with heterotopic pregnancies, particularly in patients with a history of salpingectomy. Identifying these factors can help in developing preventive strategies and risk assessment tools. Investigating advanced diagnostic techniques and imaging modalities could enhance early detection and accurate diagnosis of heterotopic pregnancies, providing critical insights into the efficacy of different diagnostic tools in varied clinical scenarios.

Developing and refining management protocols for heterotopic pregnancies can improve patient outcomes, with studies comparing different surgical and medical treatment options offering valuable information on the most effective approaches for managing such complex cases. Longitudinal studies following patients with heterotopic pregnancies and their offspring can provide essential data on the long-term outcomes and any potential complications associated with this condition, guiding postnatal care and support.

This case contributed to the growing body of literature on heterotopic pregnancy and showed the need for ongoing research and clinical vigilance to improve diagnosis, management, and outcomes for such patients. Through addressing these clinical and research implications, healthcare providers can better navigate the complexities of heterotopic pregnancies and enhance care for future patients facing similar challenges.

Ethical Considerations

In this case, explicit consent from the patient for publication was not acquired. Nonetheless, the data were handled and processed with utmost care to protect the patient's anonymity, ensuring that no identifiable information was included. We adhered strictly to all ethical protocols regarding confidentiality, and all personal details were omitted to preserve the patient's privacy. The preparation and reporting of this case followed the ethical guidelines established by the hospital's ethics committee.

Data availability

The data supporting the findings of this case report can be obtained from the corresponding author upon reasonable request. Clinical information, including diagnostic tests, medical history, and treatment details, were gathered and analyzed in accordance with institutional privacy policies and ethical standards. However, to safeguard patient confidentiality, certain personally identifiable information has been removed in compliance with privacy and ethical considerations.

Acknowledgments

The authors acknowledge the contributions of the healthcare team involved in the patient's care, including the surgical and nursing staff who ensured the patient's well-being throughout her treatment. Their professional expertise and compassionate care were instrumental in managing this complex case successfully.

Author Contributions

Each author contributed significantly to the conception, design, and writing of this case report. Each author

reviewed and approved the final manuscript.

Conflicts of Interest

The authors declare no conflicts of interest. There were no financial or personal relationships that could have inappropriately influenced the work reported in this paper.

Funding Sources

No specific funding was received for the creation of this case report. The publication fees were supported by the authors themselves, and there were no external funding sources involved.

References

1. Johnson NP, Mak W, Sowter MC. Surgical treatment for tubal disease in women due to undergo in vitro fertilisation. *Cochrane Database Syst Rev.* 2004;(3):CD002125. doi: 10.1002/14651858.CD002125.pub2. Update in: *Cochrane Database Syst Rev.* 2010 Jan 20;(1):CD002125. doi: 10.1002/14651858.CD002125.pub3. PMID: 15266464.
2. Dor J, Seidman DS, Levran D, Ben-Refael Z, Ben-Shlomo I, Mashiach S. The incidence of combined intrauterine and extra uterine pregnancy after in-vivo fertilization and embryo transfer. *Fertil Steril.* 1991;55:833-4.
3. Shavit T, Paz-Shalom E, Lachman E, Fainaru O, Ellenbogen A. Unusual case of recurrent heterotopic pregnancy after bilateral salpingectomy and literature review. *Reprod Biomed Online.* 2013;26:59-61.
4. Barrenetxea G, Barinaga-Rementeria L, Lopez de Larruzea A, Agirregoikoa JA, Mandiola M, Carbonero K. Heterotopic pregnancy: two cases and a comparative review. *Fertil Steril.* 2007;87(2):417e9-417e15.
5. Clayton H, Schieve L, Peterson H, Jameison D, Reynolds M, Wright V. A comparison of heterotopic and intrauterine-only pregnancy outcomes after assisted reproductive technologies in the United States from 1999 to 2002. *Fertil Steril.* 2007;87:303-9.
6. Talbot K, Simpson R, Price N, Jackson SR. Heterotopic pregnancy. *J Obstet Gynaecol.* 2011;31:7-12.
7. Lim L, Fuentes H. Spontaneous intrauterine pregnancy after tubal sterilization: A case report. *SAGE Open Med Case Rep.* 2024;12:1-3. <https://doi.org/10.1177/2050313X241251732>
8. Baltus T, Brown J, Molakatalla S, et al. Spontaneous pregnancy after total bilateral salpingectomy: a systematic review of literature. *J Minim Invasive Gynecol.* 2022;29:213-8.
9. Bollapragada SS, Bandyopadhyay S, Serle E, et al. Spontaneous pregnancy after bilateral salpingectomy. *Fertil Steril.* 2005;83:767-8.
10. Mills K, Marchand G, Sainz K, et al. Salpingectomy vs tubal ligation for sterilization: a systematic review

and meta-analysis. *Am J Obstet Gynecol.* 2021;224:258.e4-265.e4.

11. Clark NV, Endicott SP, Jorgensen EM, et al. Review of sterilization techniques and clinical updates. *J Minim Invasive Gynecol.* 2018;25:1157-64.
12. Kim AJ, Barberio A, Berens P, et al. The trend, feasibility, and safety of salpingectomy as a form of permanent sterilization. *J Minim Invasive Gynecol.* 2019;26:1363-8.
13. Peterson HB, Xia Z, Hughes JM, et al. The risk of pregnancy after tubal sterilization: findings from the U.S. collaborative review of sterilization. *Am J Obstet Gynecol.* 1996;174:1161-8.
14. Mowat A, Maher C and Ballard E. Surgical outcomes for low-volume vs high-volume surgeons in gynecology surgery: a systematic review and meta-analysis. *Am J Obstet Gynecol* 2016; 215: 21–33.
15. Glaser LM, King LP, Brennan L, et al. Surgeon volume in gynecologic surgery: a review of outcomes, surgical route, operative time, and cost. *J Minim Invasive Gynecol* 2018; 25: S184–S185.



Medtronic