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A Case of Ectopic Tubal Pregnancy Eight Years After a Hysterectomy Presenting as a Diagnostic Challenge

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Patient: Female, 28
Final Diagnosis: Tubal ectopic pregnancy
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: —
Specialty: Obstetrics and Gynecology

Objective: Unusual clinical course
Background: Ectopic pregnancy after hysterectomy is extremely rare, and the diagnosis can be challenging, even with modern imaging methods. A rare case is presented of ectopic tubal pregnancy in a 28-year-old woman that presented eight years following a hysterectomy.

Case Report: A 28-year-old woman was admitted in our hospital with moderate hypogastric pain, mild vaginal bleeding, and bilateral mastalgia. She had undergone hysterectomy eight years previously for postpartum hemorrhage. Pelvic ultrasound showed a heterogeneous tissue mass in the pelvis with peripheral vascularity that was also seen on magnetic resonance imaging (MRI). Laboratory tests showed increased serum levels of human chorionic gonadotropin (hCG). Following hospital admission, she developed an acute abdomen and shock due to tubal rupture and an emergency salpingectomy was performed.

Conclusions: A multidisciplinary approach to diagnosis and management facilitated an accurate and timely diagnosis in a rare case of ectopic pregnancy that presented eight years following hysterectomy, but could not prevent the development of potentially life-threatening complications.

MeSH Keywords: Hysterectomy • Magnetic Resonance Imaging • Pregnancy, Ectopic • Pregnancy, Tubal • Ultrasonography

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Background

Ectopic pregnancy after hysterectomy is an unexpected event that was first reported by Wendler in 1895 [1]. Currently, 73 cases of this rare condition have previously been reported, with the last case being reported by Ahmed et al. in May 2019 [2]. Ectopic tubal pregnancy after hysterectomy includes an early presentation in the immediate period after hysterectomy, which represents most of the reported cases, and late presentation from seven months to 12 years after hysterectomy [1,3]. In cases of early presentation, an undiagnosed pregnancy existed at the time of hysterectomy [1,4].

A case is reported of ectopic tubal pregnancy in a 28-year-old woman, eight years after hysterectomy. To our knowledge, this is the 74th reported case of ectopic pregnancy following previous hysterectomy.

Case Report

A 28-year-old woman (gravida 5, para 2) presented to our Emergency Department with a one-week history of moderate hypogastric pain, mild vaginal bleeding, and bilateral mastalgia. According to her medical records, eight years previously, she had a history of a cesarean section for fetal distress and fetopelvic disproportion during her fifth pregnancy, followed by total hysterectomy for postpartum hemorrhage. No immediate or long-term complications had been reported after surgery.

On admission to our emergency department, physical examination showed normal vital signs. The patient had mild discomfort and pain on deep palpation of the left lower abdominal quadrant. Routine pelvic examination identified congested vaginal mucosa and bleeding from a small circular hole measuring approximately 2 mm in the vaginal vault scar line, but no cervix was seen. Ultrasound imaging of the breasts was normal with no cysts or masses.

Transvaginal ultrasound initially showed a heterogeneous and well-defined pelvic mass, with multiple cystic spaces, which was in direct contact with the vagina and bladder (Figure 1). There was also another heterogeneous tissue mass on the left side that was poorly defined and had a central intralesional cystic mass surrounded by a hyperechogenic halo (Figure 2). The cystic mass showed intense peripheral vascularity with a 'ring of fire' appearance, indicating a high pulsatility index (Figure 3). The pelvic ultrasound report confirmed the presence of fluid in the pouch of Douglas.

A pregnancy test was not performed on initial hospital admission, and the patient was admitted to our department with a presumptive diagnosis of a pelvic tumor. To investigate this

Figure 1. Transvaginal ultrasound from a 28-year-old woman with a left ectopic tubal pregnancy eight years after hysterectomy. Transvaginal ultrasound shows a heterogeneous well-defined pelvic tissue mass, with multiple cystic structures in direct contact with the vagina and bladder without intralesional vascularization.

Figure 2. Transvaginal ultrasound from a 28-year-old woman with a left ectopic tubal pregnancy eight years after hysterectomy. Transvaginal ultrasound shows a left heterogeneous pelvic mass with a central cystic mass surrounded by a hyperechogenic halo.

provisional diagnosis further, serum human chorionic gonadotropin (hCG) levels (normal, <10 mIU/mL) were measured at 2 hours (928 mIU/mL) and 48 hours (1461 mIU/mL) after hospital admission. Prolactin levels were within the normal range. Magnetic resonance imaging (MRI) using a short T1 inversion recovery (STIR) sequence showed ectopic pregnancy in the ampulla of the left Fallopian tube and a residual portion of cervix in the vaginal cuff (Figure 4). Therefore, single-dose methotrexate treatment was initiated. Thirty-six hours after methotrexate administration, the patient showed signs of acute abdomen and shock, with tachycardia and hypotension, requiring

Figure 3. Transvaginal ultrasound from a 28-year-old woman with a left ectopic tubal pregnancy eight years after hysterectomy. Transvaginal ultrasound shows increased blood flow surrounding the left ectopic pregnancy with a 'ring of fire' appearance, indicating a high pulsatility index.

emergency bilateral salpingectomy. Her postoperative outcome was favorable, and the serum hCG levels decreased to 86 mIU/mL on the third day after surgery.

Discussion

Ectopic pregnancy after hysterectomy is extremely rare, and although the pathophysiological mechanism has not been

fully elucidated, the cause may be due to communication between the vagina and the peritoneal cavity, or vagino-peritoneal fistula. Several factors, such as vaginal cuff infection or vaginal cuff granulation tissue, pelvic hematoma, and transvaginal drainage of the peritoneal cavity, which are associated with Fallopian tube prolapse into the vaginal cuff, may lead to the occurrence of ectopic tubal pregnancy after hysterectomy [5–8]. Although ectopic pregnancy appears to be more common after vaginal hysterectomy (50–70% of cases), it can also occur after abdominal hysterectomy [1,5], depending on the surgical technique used.

As this case has shown, the clinical features may be nonspecific, which is why a pregnancy test was not performed on admission, resulting in a delayed diagnosis. Also, the presence of bilateral mastalgia and abdominal tenderness did not raise the possible diagnosis of ectopic pregnancy. Physical examination of the breast and ultrasound excluded the main causes of noncyclic mastalgia, such as trauma, mastitis, cysts, fibroadenoma, adenosis, and cancer, all of which rarely present bilaterally. Also, the differential diagnosis of chronic pelvic pain in an adult woman with a history of hysterectomy can be challenging, especially if it is associated with vaginal bleeding. The initial differential diagnoses that should be excluded also include gastrointestinal diseases that include appendicitis, bowel obstruction, diverticulitis, gastritis, irritable bowel syndrome, and mesenteric venous thrombosis, and urinary tract diseases, including cystitis, and pyelonephritis. The diagnosis of ectopic pregnancy is more likely to be made in cases with an early presentation in the immediate period after hysterectomy,

Figure 4. Magnetic resonance imaging (MRI) from a 28-year-old woman with a left ectopic tubal pregnancy eight years after hysterectomy. The short T1 inversion recovery (STIR) sequence on magnetic resonance imaging (MRI) shows the ectopic pregnancy in the ampulla of the left Fallopian tube (arrow) and a residual portion of cervix in the vaginal cuff.

but this was a case with a late presentation, which is much rarer [1,3]. However, as this case has shown, even in late cases of ectopic pregnancy, measurement of serum levels of human chorionic gonadotropin (hCG) are important to make the diagnosis, although some malignant tumors can be associated with increased serum hCG, including hepatic, gastric, pancreatic, ovarian, and breast cancer as well as neuroendocrine tumors. The detection of a slowly rising hCG level may be more diagnostic for ectopic pregnancy. This case has shown that a multidisciplinary approach to the diagnosis of ectopic pregnancy also included imaging, which identified the location of the fetus and fetal sac in the antrum of the left Fallopian tube.

This patient was initially offered laparoscopic surgery for both diagnosis and treatment, but the patient initially refused surgery and chose conservative treatment with methotrexate. Methotrexate treatment in ectopic pregnancy after hysterectomy has not been well documented. Following the start of methotrexate treatment, the patient developed hemorrhagic shock that required emergency laparoscopic surgery. At surgery, acute intra-abdominal hemorrhage and pelvic adhesions were present, and because the source of bleeding could not be identified, laparotomy and adhesiolysis were performed to identify a ruptured left-sided ectopic tubal pregnancy, in close contact with a small fragment of residual cervix, which was confirmed by pathology examination. These findings confirmed the association between residual cervix and ectopic tubal pregnancy following a hysterectomy.

In this case, left salpingectomy was performed with prophylactic right salpingectomy, but the residual cervix was not removed due to the extensive adhesions and the risk of causing a vesicovaginal fistula, which is a common complication in such cases. In some circumstances, the residual cervix can be cauterized to prevent the formation of a vagino-peritoneal fistula, which was not considered in this case. When the cervix remains *in situ*, techniques should be used to obliterate or isolate the residual cervical canal [1]. In this case, covering the cervical stump with peritoneum was also a possible surgical approach.

The use of prophylactic bilateral salpingectomy at the time of hysterectomy to prevent ectopic pregnancy remains controversial, as does whether bilateral salpingectomy affects ovarian viability. In this case, even though a previous laparoscopic total hysterectomy was performed eight years previously, the persistence of a residual portion of the cervix in the vaginal

cuff made possible the formation of a vagino-peritoneal fistula which facilitated the migration of sperm [9,10]. Emergency hysterectomy for hemorrhage that includes dilatation of the cervix can result in lack of epithelialization of the vaginal vault due to tissue hypoperfusion, which facilitates the formation of a vagino-peritoneal fistula, with or without accidental persistence of a residual cervix [1,10,11]. The method of vaginal cuff closure differs between different types of hysterectomy, performed vaginally or abdominally [1,3]. Also, as in the case, the Fallopian tube can be in close contact with the residual cervix, which was another factor that favored ectopic pregnancy. For this reason, it is very important that vaginal cuff closure during hysterectomy does not incorporate the Fallopian tubes, which can depend on the experience and training of the surgeon and the surgical technique used [1,2,12].

Conclusions

A rare case is presented of a 28-year-old woman with ectopic left tubal pregnancy that presented eight years following a hysterectomy, possibly due to the close contact of the Fallopian tube with residual cervix and the formation of a vagino-peritoneal fistula. This case has shown that it is important to be aware that a young woman with a history of hysterectomy can develop ectopic pregnancy. The type of hysterectomy and the operative technique used are important risk factors for this rare event. Even if the clinical symptoms and signs are nonspecific, measurement of serum human chorionic gonadotropin (hCG) levels can raise the index of suspicion for ectopic pregnancy after hysterectomy. Ultrasound combined with magnetic resonance imaging (MRI) can provide a diagnosis as rapidly as possible as delay in the diagnosis of this rare condition may lead to potentially fatal complications. However, prophylactic bilateral salpingectomy at the time of hysterectomy to prevent ectopic pregnancy remains controversial.

Acknowledgments

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Conflict of interest

None.

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