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A Rare Case of Abdominally Placed, Non-communicating Tubal Pregnancy with Unicornuate Uterus

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Abstract

This case report aims to present a very rare case of abdominal pregnancy localized in the non-communicating fallopian tube in a patient with a unicornuate uterus. A 32-year-old patient presented with abdominal pain and high beta-human chorionic gonadotrophin levels and was suspected of ectopic pregnancy. Diagnostic laparoscopy revealed unicornuate uterus with a normal fallopian tube and ovary on the left side. On the contralateral side, right ovary and fallopian tube were explored adherent to the abdominal wall as well as the tubal ectopic focus in the non-communicating fallopian tube. Then, salpingectomy was performed. The female reproductive system was formed by the migration and fusion of bilateral Müllerian ducts. Due to the incomplete transabdominal descent of the ovary, the ipsilateral Müllerian duct could not complete its caudomedial development and failed to fuse and form the uterovaginal primordium in this patient, resulting in a unicornuate uterus and an abdominal ovary and fallopian tube. Interestingly, ectopic pregnancy occurred in the abdominally placed non-communicating fallopian tube in this case. To our knowledge, this is the first case to report an ectopic pregnancy that occurred in an abdominally placed non-communicating fallopian tube in a patient with unicornuate uterus. Ectopic pregnancy is an important complication of Mullerian duct anomalies and should be diagnosed and treated early by laparoscopy.

Keywords: Abdominal pregnancy, Mullerian ducts, fallopian tube

Introduction

The implantation of the embryo outside the normal uterine cavity is defined as ectopic pregnancy.[1] Ectopic pregnancy may be observed in cervix, ovary, myometrium, abdominal cavity, and most predominantly in the fallopian tube. The prevalence of ectopic pregnancy is 1%-2% in the general population but increases up to 5% in patients with assisted reproductive technology. Even though the mortality decreased due to early diagnosis with ultrasound and serial serum beta-human chorionic gonadotrophin (β-hCG) measurements, the ruptured ectopic pregnancy is still a major cause of maternal mortality and morbidity.[2] Age, history of ectopic pregnancy, tubal operation or tubal damage, pelvic inflammatory disease, assisted reproductive technology, intrauterine device, and smoking are identified as risk factors for ectopic pregnancy.[1] Another rare but significant risk factor for ectopic pregnancy is the Müllerian anomaly. Congenital anomalies of the uterus may present with variable complications, including ectopic pregnancy.[3,4]

In the literature, there were cases presented with abdominal ectopic pregnancy which caused serious complications such as massive hemorrhage and bowel injury, being attached to the uterine wall, bowel, mesentery, liver, spleen, bladder, and ligaments.[5-7] To our knowledge, this is the first case to report an ectopic pregnancy that occurred in an abdominally placed non-communicating fallopian tube in a patient with unicornuate uterus, diagnosed and treated with laparoscopic surgery. This case emphasizes a unique embryologic anomaly that occurred due to incomplete migration of the Mullerian duct during embryogenesis. This case report was prepared according to CARE case report guidelines.

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Case Presentation

A 32-year-old-woman with a complaint of abdominal pain for 2 days presented to our outpatient clinic. Her last menstrual period date was unknown. She had epilepsy and urinary tract anomalies which were right renal agenesis, left pelvic kidney, cross-fused renal ectopia, and duplicated collected system anomaly. Her urinary tract anomalies were diagnosed at the age of 8 with the complaint of recurrent urinary tract infection. She had inguinal hernia operation, cystoscopy, and left myringotomy. For epilepsy which was diagnosed at the age of 16, she used sodium valproate 250 mg/day and methyldopa for blood pressure control.

On the day of her admission, her physical examination revealed neither abdominal defense nor rebound. In the transvaginal ultrasound examination, the endometrium measured 5 mm and the free fluid collection was observed in rectouterine pouch. The serum β-hCG level in her blood test was 844.2 mIU/mL. She was hospitalized for a follow-up of her symptoms, total blood count, and serum β-hCG levels. Methotrexate was not advised by internal medicine specialist due to its nephrotoxicity. The follow-up of hemoglobin, hematocrit, and β-hCG levels of the patient was summarized in Figure 1. On the eighth day, reduction of hemoglobin and serum β-hCG levels became evident with newly onset abdominal tenderness and rising amount of free pelvic fluid. In the transvaginal ultrasound examination, the endometrium measured 14 mm, and

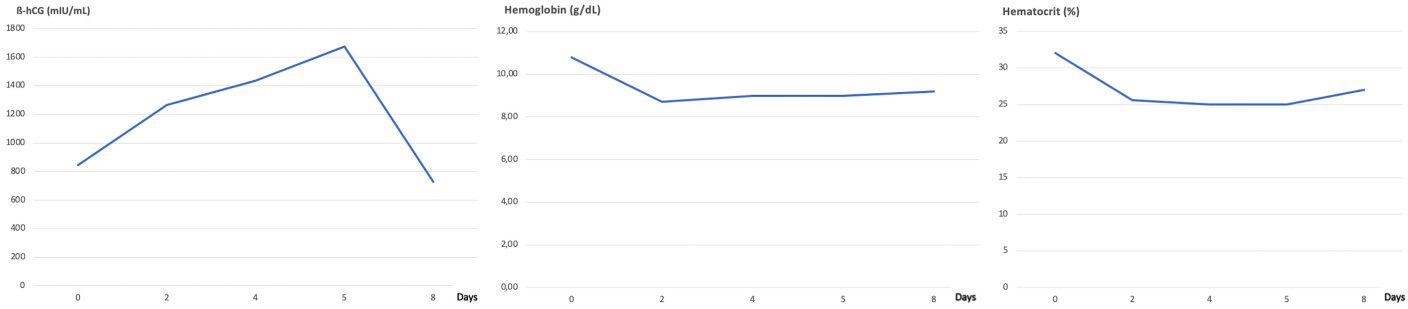

Figure 1. Serum β-hCG levels and hemoglobin and hematocrit follow-up of the patient. β-hCG, beta-human chorionic gonadotropin.

heterogenous fluid was observed in the endometrial cavity. The free pelvic fluid in the pelvic region increased. Diagnostic laparoscopy decision was made.

Entrance to the abdominal cavity was done from the umbilical region with a closed technique (with Veress needle). The abdomen was inflated with 3 L of carbon dioxide. Trocars of 10 mm were inserted from the umbilicus and left lower quadrant. The hemorrhagic fluid in the rectouterine pouch was aspirated and the unicornuate uterus, left fallopian tube, and ovary were seen normal in size and shape (Figure 2). The right ovary and fallopian tube were not observed in the right adnexal region. There was no ectopic focus identified in the pelvic region. Further exploration revealed an ectopic ovary adherent to the right abdominal wall, near the lumbar region. There was a hemorrhagic tubal ectopic focus in the non-communicating fallopian tube adjacent to the ovary (Figure 2). Salpingectomy was performed with the help of bipolar electrocautery. After bleeding control, the operation was completed. The patient was discharged from the hospital on the second day after the operation without any postoperative complications. The pathology of the surgical specimen was reported as trophoblastic cells in the coagulum in the tubal wall (ectopic pregnancy).

Discussion

This case presents a very rare ectopic pregnancy that occurred in an abdominally placed non-communicating fallopian tube in a patient with a unicornuate uterus. It emphasized the importance of embryological knowledge in the differential diagnosis of ectopic pregnancy and the importance of diagnostic laparoscopy. Müllerian duct anomalies are generally asymptomatic at the early stages of life, but ectopic pregnancy may be an important clinical presentation of Müllerian anomalies due to its morbidity and mortality.

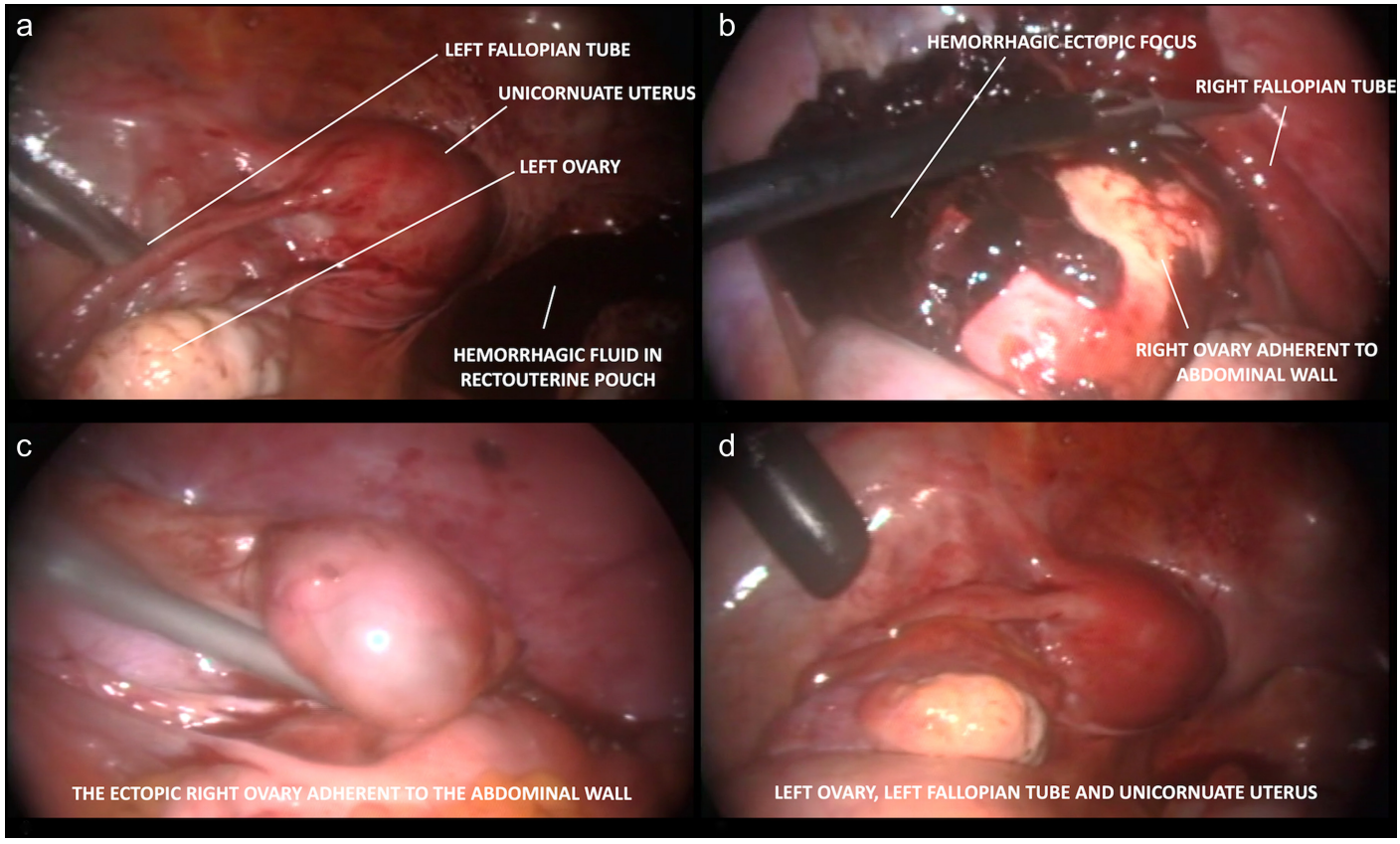

Figure 2. a-d. Laparoscopic surgery. (a).Unicornuate uterus, left fallopian tube, and left ovary with hemorrhagic fluid in rectouterine pouch. (b) Ectopical right ovary and fallopian tube with hemorrhagic ectopic focus on right abdominal wall. (c) Ectopic right ovary after salpingectomy. (d) Unicornuate uterus, fallopian tube, and ovary at the end of surgery.

In this case, we presented an anatomical variant of Müllerian duct derivatives: a unicornuate uterus with an abdominal non-communicating fallopian tube and ovary which can not be classified according to The American Fertility Society Müllerian Anomaly Classifications.[8] During embryogenesis, Müllerian ducts appear at sixth to seventh weeks of pregnancy as an invagination of coelomic epithelium in the lateral wall of the cranial end of urogenital ridge, adjacent to the mesonephric duct.[9] The free edges unite to form the duct except at the site of ostium of the fallopian tube. After the caudomedial growth of Müllerian ducts, around the eighth week, the caudal parts of contralateral ducts fuse and the septum degenerates to form a single cavity (uterovaginal primordium) (Figure 3). The non-fused part of each Mullerian duct becomes the fallopian tube, and the distal end remains open as the ostium. Normally, the gubernaculum leads the transabdominal "descent" of the ovary, but this descent is restricted due to the attachment of the gubernaculum to the paramesonephric duct.[9,10] But, in this case, probably due to the incomplete descent of ovary, the ipsilateral Müllerian duct could not complete its caudomedial development and failed to fuse and form uterovaginal primordium. As a result of this, the patient has unicornuate uterus and abdominally placed ovary and non-communicating fallopian tube. In the literature, there are other cases of undescended ovary diagnosed in childhood period with acute abdominal pain who also had genitourinary abnormalities such as unicornuate uterus or renal agenesis diagnosed by laparoscopy or magnetic resonance imaging like in our case.[11-14]

Congenital anomalies of the uterus result from arrested development, abnormal formation, or incomplete fusion of Müllerian ducts with a prevalence of 0.1%-3.8% in the general population and 6.3% in the infertile population.[15-18] Because of the complicated classification of these anomalies, diagnostic error, and delayed occurrence, the real prevalence of this anomaly is unknown.[18,19] Due to their simultaneous development and complex interactions, it should be kept in mind that abnormalities in the genitourinary system are common among patients with Müllerian anomalies with approximately 40% of patients having coexisting renal abnormalities.[15] Supporting this information, the case that we presented had right renal agenesis which was the ipsilateral site of the rudimentary horn, as expected. Magnetic resonance imaging might have been considered for the anatomic configuration of uterine malformation, assessment of gestational sac, and visualization of urinary system malformations.

Regarding the pathophysiology of the ectopic abdominal pregnancy, Berghella and Wolf's[20] hypothesis about the ovum escaping fimbria trapping and getting fertilized in the abdomen has been unproven due to the failure of neovascularization shown at the implantation site of primary omental pregnancy. However, Chang et al[21] and Chopra et al[22] reported the implantation on the omentum with trophoblastic invasion and placental site reaction. Other hypotheses include delayed ovulation reversing the fertilized ovum by retrograde menstrual flow and probable fertilization occurring in the posterior cul-de-sac due to flow of peritoneal fluid or being carried to different intraperitoneal sites. Ectopic abdominal pregnancy, previously reported in the literature, occurred in the cul-de-sac, retroperitoneal or abdominal, omental region, after spontaneous or ART pregnancies[7] Placenta may be attached to the mesentery of sigmoid colon, pelvic wall, or peritoneal surface of the uterus. Similar to the cases in the literature, salpingectomy was preferred in order to decrease the further risk of ectopic pregnancies.

Diagnostic laparoscopy was an important method for the early diagnosis and treatment of a pregnancy of an unknown location

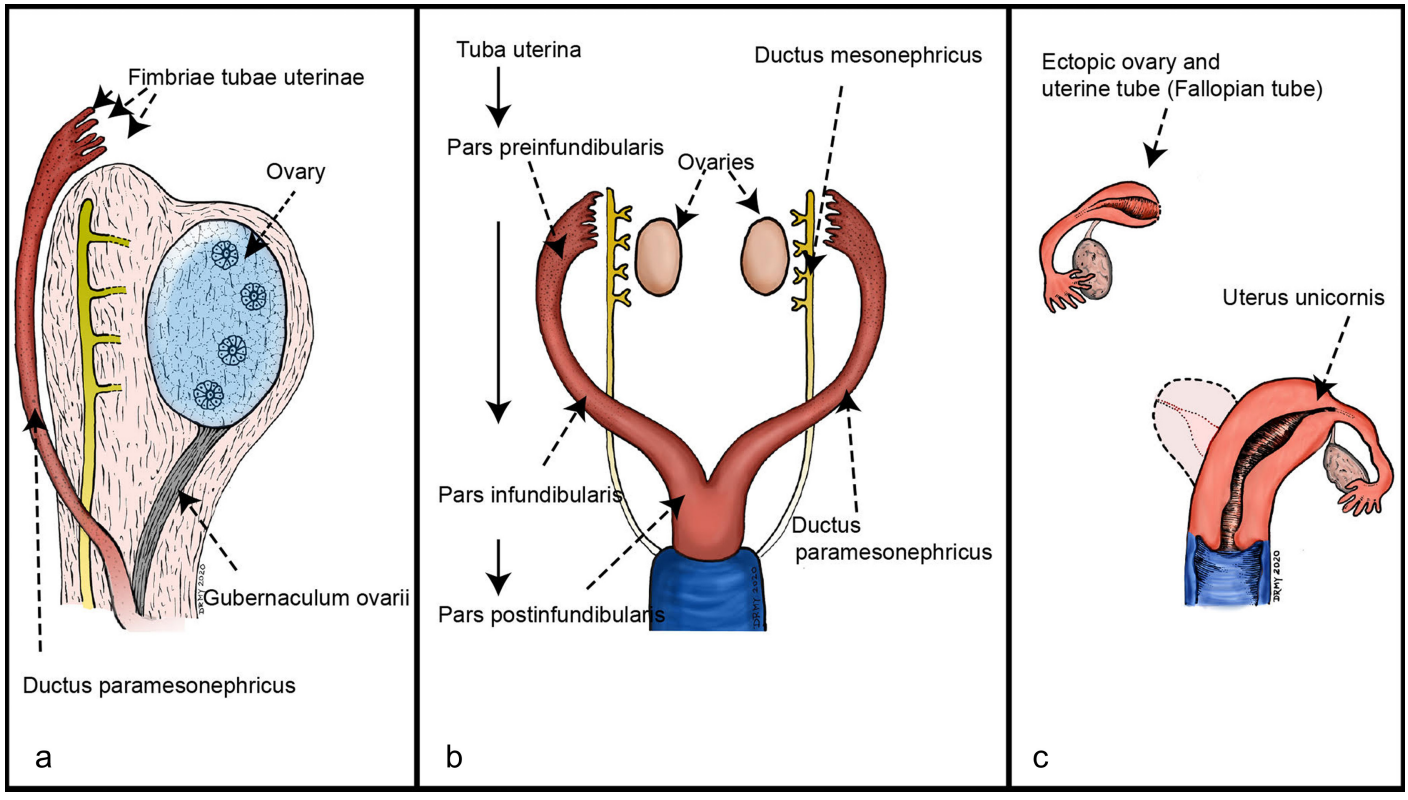

Figure 3. a-c. Anatomic illustration of Müllerian duct derivatives' development and the variation observed in the patient. (a) Ovary, gubernaculum, and Müllerian duct development. (b) Müllerian duct and its derivatives. (c) Ectopically placed ovary and fallopian tube with normally placed unicornuate uterus.

and also for identifying anatomic anomalies. When the gestational sac cannot be visualized by the transvaginal ultrasound with favorable levels of serum β-hCG, ectopic pregnancy should be excluded with additional diagnostic techniques.

Informed Consent: The patients' informed consent was taken for the performance of surgery and for the record and publication of her surgical video anonymously.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – İ.Ç.; Design – İ.Ç.; Supervision – İ.Ç.; Resources – R.M.; Data Collection and/or Processing – İ.B.Ö., M.Y., Ö.I.D.; Analysis and/or Interpretation – İ.Ç., R.M., M.Y.; Literature Search – İ.B.Ö.; Writing Manuscript – İ.B.Ö., E.B., Ö.I.D.; Critical Review – İ.Ç.

Declaration of Interests: The authors have no conflicts of interest to declare.

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