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Rare forms of ectopic pregnancy in the rudimentary horn and ovary. How not to miss the main thing?

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ABSTRACT

This article presents two cases of ectopic pregnancy in the ovary and rudimentary horn, which had a life-threatening course. Despite availability of modern high-quality diagnostics, determining the condition remains challenging, because the definitive diagnosis is established and confirmed only intraoperatively. Hence, the clinical picture can be used primarily in making a diagnosis and choosing an appropriate treatment strategy. Rare localizations, such as in the ovary and rudimentary horn, which cause difficulties in making a diagnosis, should not be overlooked. If an ectopic pregnancy is not detected promptly and the risks are underestimated, death may occur. Two clinical cases clearly demonstrate timely diagnosis, regardless of additional examination methods (e.g., pelvic ultrasound, human chorionic gonadotropin test), and adequate choice of access and scope of surgical intervention. Moreover, the lack of awareness among women about this type of pathology should be considered. Patients often do not complain, do not remember the date of their last menstruation, do not monitor their menstrual cycle, and do not seek medical help in a timely manner. The combination of several factors can lead to fatal events. In this study, both cases of ectopic pregnancy with a rare localization ended favorably.

Keywords: ectopic pregnancy; clinical case; ovarian pregnancy; pregnancy in the rudimentary horn; fetal egg.

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Редкие формы эктопической беременности: в рудиментарном роге, яичниковая беременность. Как не пропустить главное?

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АННОТАЦИЯ

В данной статье представлены два клинических случая внематочной беременности редких локализаций (яичниковой и в рудиментарном роге), которые имели жизнеугрожающее течение. Несмотря на современную качественную диагностику, широкодоступную женщинам, мы видим, что постановка диагноза вызвала затруднения, поскольку окончательно его установили и подтвердили только интраоперационно. Исходя из этого, мы сделали вывод, что клиническая картина может стать ведущей в постановке правильного диагноза и выборе правильной тактики врачом. Не следует упускать из виду такие редкие локализации, вызывающие затруднения при постановке диагноза. Если своевременно не выявить внематочную беременность и недооценить риски, то возможно развитие летального исхода. Два клинических случая ярко демонстрируют своевременную постановку диагноза, вне зависимости от дополнительных методов обследования (УЗИ органов малого таза, β-ХГЧ), и адекватный выбор доступа и объёма оперативного вмешательства. Также не следует забывать о недостаточной информированности женщин о данном виде патологии. Часто пациентки не предъявляют жалоб, не помнят дату последней менструации или вовсе не следят за менструальным циклом, несвоевременно обращаются за медицинской помощью. Сочетание нескольких факторов может привести к фатальному развитию событий. В данной статье оба случая эктопической беременности с редкой локализацией закончились благополучно.

Ключевые слова: эктопическая беременность; клинический случай; яичниковая беременность; беременность в рудиментарном роге; плодное яйцо.

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异位妊娠的罕见形式：胚角妊娠、卵巢妊娠。如何避免错过重要的事情？

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摘要

本文介绍了两个临床病例，这两个病例均为罕见的异位妊娠（卵巢妊娠和胚角妊娠），其病程危及生命。尽管现代高质量的诊断技术已广泛应用于妇女，但我们看到诊断的建立造成了困难，因为只有在术中才能最终确定和证实。在此基础上，我们得出结论，临床表现是医生做出正确诊断和选择正确策略的首要因素。这种导致诊断困难的罕见定位不应被忽视。如果不能及时发现异位妊娠，低估其风险，则有可能导致死亡。两例临床病例清楚地表明，无论采用其他检查方法（盆腔超声、β-hCG），还是充分选择手术干预的通路和范围，都能及时确诊。我们不应忘记，妇女对这类病变缺乏认识。患者往往没有任何主诉，不记得上次月经的日期，或者根本不关注自己的月经周期，也不及时就医。多种因素的综合作用会导致致命的事态发展。在本文中，两例罕见定位的异位妊娠均安全结束。

关键词： 异位妊娠；临床病例；卵巢妊娠；胚角妊娠；胎卵。

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INTRODUCTION

An ectopic pregnancy occurs when a fertilized egg embeds and develops outside the uterus. Ectopic pregnancy may be tubal, ovarian, within the rudimentary horn, abdominal, or combined [1]. Ectopic pregnancy was first described after the patient's death. The lifetime diagnosis of ectopic pregnancy was made in 1812.

Survival rates were low, so this condition was a focus of much research. In Russia, ectopic pregnancy was studied by Zmigrodsky, Fenomenov, Snegiryov and his students [2].

Statistics show that 1.3–2.4% of pregnancies are ectopic. The mortality rate is 7.4% [3]. The incidence of ectopic pregnancy is 1.4% in the USA and 2.0% in Germany [4].

The case fatality rate in ectopic pregnancy remains high. In Russia, this parameter was 0.47 per 100,000 live births in 2012, 0.21 in 2013, and 0.26 in 2014 [2].

The main mechanisms underlying ectopic pregnancy include abnormal transport of a fertilized egg through the fallopian tube, high trophoblast activity [3], obstruction of the fallopian tubes and changes in their walls, as well as ciliary disorders [5].

Ectopic pregnancy is usually diagnosed between 6 and 9 weeks of gestation.

Risk factors for ectopic pregnancy are as follows:

1) A high-risk group:
- History of fallopian tube surgery,
- History of ectopic pregnancy,
- Use of an intrauterine device.

2) A moderate- risk group:
- Ascending infections,
- Tobacco use,
- Fallopian tube disorders.

3) A low-risk group:
- Age over 40 years [4].

Ectopic pregnancy can be caused by inflammatory diseases (42%–80%) [2, 3]; endometriosis due to abnormal transport through the fallopian tubes with potential for their mechanical damage; obstruction [2]; abnormalities of the fallopian tubes, for example, an infantile fallopian tube (narrow, long, tortuous, scarred, etc.) [1–3]; a history of artificial abortion [1, 2]; hormonal changes [1]; lesions in the uterus and its appendages leading to abnormal organ arrangement and abnormal transport through the fallopian tubes [1–3]; in vitro fertilization, which doubles the incidence of ectopic pregnancy [2]; stress; nervous excitement leading to spasm of the fallopian tubes [1, 2].

Manifestations of ectopic pregnancy vary, ranging from asymptomatic condition to fallopian tubal rupture with hemorrhagic shock [4], which significantly impedes the diagnosis.

Abdominal pain (95%) is the most common symptom. Other symptoms include late periods delayed for a few weeks (90%) and bloody, often spotty, vaginal discharge (50–80%) [2].

The first person to identify an ectopic pregnancy by X-ray was Klaus in 1908. Before that, the diagnosis was based solely on medical history and palpation. In 1926, Dyroff proposed the use of hysterosalpingography in pregnant women up to the 12th week of gestation because it could detect uterine hypotonia. However, in 1939, after conducting a study, Schultze Gunter concluded that uterine hypotonia was observed in only 40% of pregnant women. In 1963, Weinberg et al. presented data showing that the combined use of pneumopelviography and hysterosalpingography allowed a more accurate diagnosis of ectopic pregnancy. In 1969, echography was first used by Kobayashi et al.

In 1990, in search for methods for more accurate diagnosis of ectopic pregnancy, Demidov and Zykin proposed echographic diagnostic signs:

1) The presence of a structurally complex appendage neoplasm as a result of tubal rupture or miscarriage;

2) Absence of a gestational sac in the uterine cavity;

3) The smaller uterus than expected for the gestational age (provided there are no uterine disorders);

4) Endometrial thickening;

5) Detection of a pseudo gestational sac in the uterine cavity;

6) Free fluid in the pelvic cavity.

Later, these signs were significantly revised.

In the early 1990s, transvaginal ultrasound was introduced and many authors considered this method to be highly accurate. There are modern echographic criteria for ectopic pregnancy developed by Kirk, Bottomley, and Bourne in 2014.

They include:

1) Presence of a heterogeneous extraovarian non-cystic appendage lesion caused by dilatation and alteration of the fallopian tube with formation of a detached, often blighted, egg and blood clots;

2) Detection of a spherical cavity lesion in the form of a tube with hematosalpinx as a substrate;

3) The tubal ring sign (also known as bagel sign or blob sign); and

4) Ectopic gestational sac [4].

Transvaginal ultrasonography can detect ectopic pregnancy at 1.5–3.0 weeks of gestation.

Since the mid1980s, color Doppler mapping and pulsed-wave Doppler have been used (A. Kurjak) to visualize increased vascularity in the area of the ectopic trophoblast. This is a more advanced modality to diagnose ectopic pregnancies [2].

Changes in human chorionic gonadotropin (HCG) levels can be seen as early as 7–8 days after fertilization. In case of an ectopic pregnancy, the HCG level does not increase by more than 66% or decrease by more than 13% from baseline within 48 hours. A ratio within this range with an absolute HCG level above 1,500 IU/L may suggest an ectopic pregnancy [4].

The literature shows that progesterone levels are lower in ectopic pregnancy than in intrauterine pregnancy and this

value can be used for differential diagnosis. Some evidence suggests that blood progesterone levels of less than 15 ng/mL indicate an ectopic pregnancy in 80% of cases [2].

Laparoscopy is one of the most accurate diagnostic methods with up to 100% accuracy. Laparoscopy can visually assess the condition of the uterus, ovaries, fallopian tubes, the amount of blood loss, the location of the ectopic egg, the type of pregnancy, and perform surgical treatment.

Magnetic resonance imaging (MRI) is also used to diagnose ectopic pregnancy. However, its effectiveness in early pregnancy has not yet been established. MRI during pregnancy shows a layer of normal myometrium in the rudimentary horn around the gestational sac. The dominant horn will be offset to the side and has a banana shape. An abdominal pregnancy is characterized by the absence of myometrium around the gestational sac [5].

Currently, there are several treatment options for ectopic pregnancy, including surgical, medical, and expectant management [2].

Surgical treatment options include laparoscopy and laparotomy. Advantages of laparoscopy include faster access to the abdominal cavity, less intraoperative trauma, less blood loss, and less potential for postoperative adhesions [1, 4, 6]. Laparoscopy is used for both radical (salpingectomy) and conservative plastic surgery. Laparoscopy is performed when the patient's condition is satisfactory and hemodynamics are stable. Grade 3–4 hemorrhagic shock is an absolute contraindication to laparoscopy. Relative contraindications include hemodynamic instability (grade 1–2 hemorrhagic shock); interstitial location of the gestational sac; location of the gestational sac in the accessory uterine horn; rupture of the fallopian tube wall. General contraindications to laparoscopy include obesity, severe adhesions, and cardiovascular and pulmonary failure [2].

Hysterofibroscopy is another modality which is performed only in early pregnancy, up to four weeks of gestation [1].

The literature provides data on the use of methotrexate, an anti-tumor agent, as a non-surgical treatment. Methotrexate is a structural analogue of folic acid which prevents folic acid from being converted to its active form and inhibits the synthesis of amino acids necessary for development of embryonic DNA [2]. According to various literature data, the success rate of methotrexate treatment varies from 63% to 97%. Methotrexate has some side effects such as nausea, vomiting, stomatitis, diarrhea, high liver enzymes, kidney and liver damage, pneumonia, dermatitis, and pleurisy [4]. Methotrexate can be administered systemically (orally and parentally), locally, and transcervically during laparoscopy. Combined use is also possible. However, due to many side effects, this agent is currently not used in clinical practice.

Less commonly, potassium chloride and hypertonic dextrose are used for non-surgical treatment [2].

Some authors report that one in four patients experience recurrent ectopic pregnancy, one in five to six develop pelvic adhesions, and 75% experience secondary infertility after tubectomy [3].

Rare types of ectopic pregnancy include ovarian (1.0–3.0%), cervical (0.1–0.4%), abdominal (0.1–1.4%), intraligamentary (0.1%), and rudimentary horn (0.1–0.9%) pregnancies.

A rudimentary horn pregnancy is an extremely rare type of ectopic pregnancy. According to Radzinsky, its incidence is 0.1–0.9% [7]. Global literature estimates the incidence of rudimentary horn pregnancy to be at 1 in 100,000–140,000 [8]. These uterine anomalies are caused by developmental defects in the Mullerian duct. Unilateral ductal hypoplasia results in a unicornuate uterus with a rudimentary horn. Rudimentary horn pregnancy may develop due to transperitoneal migration of spermatozoon or a fertilized egg followed by implantation into the horn [9].

Due to the thinning of the rudimentary horn endometrium, the myometrium is less vascularized and the connective tissue component predominates, leading to high risk of placenta accreta spectrum with subsequent uterine rupture. In most cases, this occurs between 8 and 16 weeks of gestation [7].

Nahum estimates that 5.3% of all pregnancies are double pregnancies, one in the rudimentary horn and the other in the functional horn [8].

In addition to uterine rupture, risks include infertility, severe fetoplacental insufficiency leading to antenatal fetal death, or severe intrauterine growth retardation. The maternal mortality rate is 5%.

The first case of this type of pregnancy was described by Dreier in 1894. Pregnancy was located in the right rudimentary horn, and the corpus luteum was located in the left ovary [3].

Early diagnosis is challenged because this type of pregnancy is mostly asymptomatic and is often discovered incidentally during treatment for infertility, pelvic pain, miscarriage, or in the second trimester when a uterine rupture occurs [9, 10].

Ultrasound, especially transvaginal ultrasound, hysterosalpingography, computed tomography, and magnetic resonance imaging are of great importance for diagnosis. Double pregnancy can be diagnosed during laparoscopy [8]. This condition may be suspected in the presence of a soft, painless "tumor" protruding from the slightly enlarged and mobile uterus, on a thick pedicle. Echography is a highly accurate method of detecting pregnancy in a rudimentary uterine horn.

There are ultrasound criteria for a rudimentary horn pregnancy, recommended by the Royal College of Obstetrics and Gynecology (RCOG) in 2016:

1) A single interstitial portion of a fallopian tube in the main uterine body;

2) A mobile gestational sac surrounded by myometrium but separated from the uterus; and

3) A vascular pedicle adjoining the gestational sac to the unicornuate uterus [11].

Additional ultrasound criteria include the distance from the center of the gestational sac to the side wall of the uterus greater than 1 cm and the thickness of the myometrium surrounding the gestational sac less than 5 mm.

There are data on full-term pregnancies with the gestational sac in the rudimentary horn and data on fetal death only at 34 weeks of gestation [7].

A treatment strategy for rudimentary horn pregnancy includes removal of the horn with the ovum by laparoscopy or laparotomy in combination with ipsilateral salpingectomy and metroplasty. The Russian Society of Obstetricians and Gynecologists recommends planning another pregnancy not earlier than 8–10 months after the failing pregnancy [11].

Ovarian pregnancy is an ectopic pregnancy with the gestational sac located in the ovary. Maternal mortality rates range from 14.3% to 50.0%. The incidence is 1–3% [12].

Ovarian pregnancy was first reported in 1682 in Saint Maurice [3].

The Spiegelberg's criteria for an ovarian pregnancy include: fallopian tubes must be intact and separate from the ovary; the gestational sac must occupy the normal position of the ovary; the gestational sac must rest on the uterus through the uteroovarian ligament; there must be ovarian tissue in the wall of the gestational sac [13].

There are several types of ovarian pregnancy:

- Intrafollicular (true, primary), where fertilization occurs within the follicle, a rare one;
- Ovarian (secondary), where the embryo is located on the surface of the ovary [12, 14]; and
- Interstitial, where the gestational sac descends toward the ovary [15].

The bilateral form is rare [16].

The causes of ovarian pregnancy are not fully understood, but the most likely causes include infectious diseases, cystic ovarian neoplasms, postoperative complications, impaired tubal transport, endocrine disorders, genital infantilism, use of an intrauterine device, endometriosis [6, 12, 14]. Many foreign sources report use of intrauterine devices as a key risk factor [6, 16].

According to some data, abnormal changes in the fallopian tubes and previous tubal surgery are not risk factors for ovarian pregnancy [16]. However, other literature reports ovarian pregnancy following tubal pregnancy and associated tubal removal [17].

In addition, ovarian pregnancy often occurs in patients after in vitro fertilization [13, 16].

Ovarian pregnancy most commonly occurs in women aged 20–34.

Symptoms include abdominal pain in 42.9% and vaginal bleeding in 28.6% [12, 14]. Abdominal pain suggests a rupture of the ovarian capsule and development of hemoperitoneum. Therefore, patients are usually admitted in shock [16]. There are data supporting the asymptomatic course of this condition [6].

Diagnostic modalities for ovarian pregnancy include bimanual vaginal examination, ultrasound, beta HCG testing, and laparoscopy.

Ultrasound criteria for ovarian pregnancy include absence of the gestational sac in the uterus and fallopian tubes; corpus luteum-like structures in the ovary; the gestational sac with peripheral vascularity (the ring of fire sign) in the projection of the affected ovary; signs of intra-abdominal bleeding; visual possibility of culdocentesis [13].

Some authors believe that MRI is an important diagnostic modality because it allows a more precise localization of the gestational sac [4]. In most cases, the condition is diagnosed during surgery [6]. Differential diagnosis includes corpus luteum cyst or cyst with hemorrhage [16].

More full-term pregnancies are reported with this type of ectopic pregnancy than with other types. For example, Dane et al. reported a case of ovarian pregnancy carried to term up to 32 weeks with a live fetus weighing 1,400 g [13]. This is possible due to a well-developed vascular network, a relatively large cavity for egg implantation within the follicle, and the ability of the ovarian tissue to proliferate rapidly.

However, pregnancy is often interrupted at the early stages, with severe abdominal pain, heavy bleeding, nausea, vomiting, dizziness, and syncope [15, 17].

Foreign studies report cases of ovarian pregnancy in elderly women [14].

Ovarian pregnancy is treated surgically (oophorectomy or wedge resection of the ovary). Laparoscopy or minilaparotomy is used [3, 6, 17].

The literature shows that in addition to these procedures, a trophoblast curettage with coagulation and ovarian conservation is also performed [16]. Before surgery, pitressin may be used to temporarily constrict blood vessels to minimize intraoperative blood loss [6]. Further non-surgical treatment with methotrexate may be considered in the event of a postoperative increase in beta-GCG levels, which may indicate deep trophoblast invasion [3, 12, 16, 17].

Two cases of rudimentary horn pregnancy and ovarian pregnancy are reported below. Informed consent for processing of personal information was obtained from the patients.

CASE DESCRIPTION

Rudimentary horn pregnancy

Patient K, 34 years old, was presented to the Emergency Department of Kaluga Regional Clinical Hospital on 5 July 2021 at 14:30.

Complaints at admission included a dragging pain in the lower abdomen that lasted for seven days.

She had been in hospital since 27 June 2021 for a threatened early abortion and received maintenance treatment (Duphaston 1 tablet twice a day). Elevated beta-HCG levels were reported, but the gestational sac was not

visualized in the uterine cavity. Due to the unclear location of the pregnancy, the patient was referred to the regional clinical hospital for further treatment.

At admission, the patient was diagnosed with a pregnancy of unknown location and mild anemia.

Her gynecologic history included periods of 5–6 days every 28 days since the age of 16 years. Periods were regular, painless, moderate. The last periods started on 17 May 2021. This was her third pregnancy (real one) after one birth and one abortion. Her past medical history included cervical ectropion. In 2007, a caesarean section was performed due to breech presentation.

The patient denied any chronic medical conditions. No history of allergy was reported. At the presentation, her condition was satisfactory. Respiratory rate was 16 breaths per minute, pulse was 72 beats per minute, blood pressure was 120/70 mm Hg. Abdomen was soft, painless, not distended. Bowel and bladder functions were normal.

Examination per vagina showed a cylindrical cervix with a closed orifice. The body of the uterus was round, soft, painless, enlarged up to 6 weeks of pregnancy. A round, soft, and painless neoplasm up to 4 cm in size was found on the left side of the uterus. Appendages on the right side were not clearly visualized and they were painless. Discharge was mucous.

Pelvic ultrasound showed the uterus to be 66 mm×52 mm×75 mm. M-echo showed a heterogeneous lesion of 15 mm in size. The right ovary was 30 mm×27 mm without abnormalities. The left ovary was 37 mm×27 mm with follicles up to 5 mm and a heterogeneous anechoic lesion of 25 mm×20 mm. On the left side there was a heterogeneous lesion of 32 mm×33 mm, a gestational sac with an embryo, crown-rump length of 9 mm, and a positive heartbeat.

A progressive left tubal pregnancy was diagnosed.

On 6 July 2021, laparoscopy was performed to remove the additional left uterine horn and the left fallopian tube.

Laparoscopy was performed using the classic intra-abdominal approach. The uterus was round, bluish-purple, 6 cm×4 cm×6 cm in size. The right appendages included the ovary and fallopian tube without any visual abnormalities. On the left side of the uterus, a round bluish-purple neoplasm of 3.0 cm × 3.0 cm × 2.5 cm was found in relation to the left lateral uterine wall. The neoplasm was considered to be a rudimentary uterine horn. A utero-ovarian ligament (the round ligament of the uterus) and the uterine end of the fallopian tube extended from the rudimentary horn. The appendages were separated with bipolar scissors and the rudimentary horn was removed with bipolar scissors. Surgical debridement, decompression, and suturing were performed.

Table 1 shows changes in the β-subunit of HCG. Table 2 shows changes in blood parameters during the follow-up period.

During the hospital stay, clinical urinalysis, blood biochemistry, and coagulation profile were within normal limits. Blood type was 0 (I), Rh factor was positive.

Histology No. 31238-48 showed chronic salpingitis with cicatricial deformation of the fallopian tube lumen, uterine pregnancy, gravida changes in the endometrium, syncytial endometritis.

The postoperative period was uneventful and the sutures were removed on day 5 with primary intention healing. The patient was discharged in satisfactory condition.

Figures 1 and 2 show images of this case.

Ovarian pregnancy

Patient B, 38 years old, was brought to the Emergency Department of Kaluga Regional Clinical Hospital at 19:20 on 13 June 2021 by the ambulance team of the Kirovsky District Central Regional Hospital with complaints of pain in the lower abdomen that appeared 3 days ago. The patient decided to take drotaverine for pain relief, but no effect was achieved. The patient took a pregnancy test herself and it was positive.

Table 1. Human chorionic gonadotropin levels of patient K.

Date of study	Result
05.07.2021	>10,000 mIU/ml
11.07.2021	1232 mIU/ml

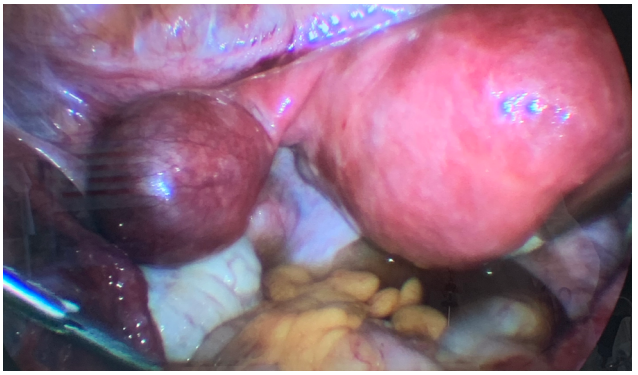

Fig. 1. Pregnancy in the rudimentary horn.

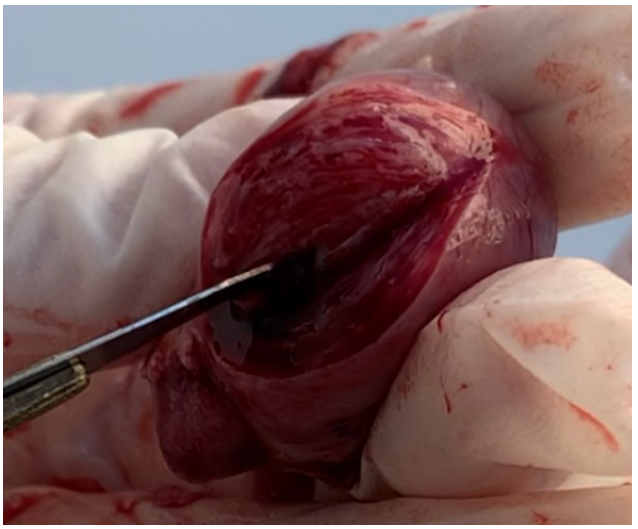

Fig. 2. Rudimentary horn.

Table 2. Clinical blood test indicators of patient K.

Date	Leukocytes, $\times 10^9$ /l	Banded neutrophils, %	Segmented neutrophils, %	Lymphocytes, %	Monocytes, %	Erythrocytes, $\times 10^{12}$/l	Hemoglobin, g/l	ESR, Mm/h	Platelets, $\times 10^9$/k
05.07.2021	9,71	7	48	32	7	4,63	116	10	221
06.07.2021	7,35	4	51	30	6	4,57	120	18	228
07.07.2021	15,14	4	86	8	2	4,99	129	16	314
10.07.2021	6,41	3	72	14	4	4,28	111	8	220

At 14:00 she went to the Central Regional Hospital where she was examined by a gynecologist and had a medical check-up (pelvic ultrasound and blood chemistry).

She reported irregular periods since April 2021, with acyclic spotting. Last period started on 17 April 2021, 3 days every 28 days. Periods were regular, painless, moderate. Two births and two abortions were reported. The patient denied chronic diseases and surgical interventions. She had a history of salpingo-oophoritis. The patient presented in moderate condition. The skin and visible mucous membranes were very pale and moderately moist. Pulse was 93 beats per minute, blood pressure was 110/70 mmHg. The abdomen was soft and very painful in all parts, not distended. Peritoneal irritation signs were positive. Bowel and bladder functions were normal.

A vaginal examination revealed the cylindrical cervix, the slit-like orifice, and extremely painful cervical traction. The body of the uterus was soft, enlarged to 7–8 weeks of pregnancy, and sensitive to palpation. On the right side of the uterus, an extremely painful volumetric mass up to 13 cm in diameter was found. The left appendage area was normal. The discharge was bloody and scanty.

Ectopic pregnancy, intra-abdominal bleeding syndrome, severe acute posthemorrhagic anemia were diagnosed preliminary.

In the Emergency Department, blood type and Rh factor were determined, a urinary catheter was placed, and fresh frozen plasma and RBCs were ordered. The patient was urgently operated. Informed voluntary consent was obtained for surgery and blood transfusion.

Considering the patient's complaints, clinical presentation, objective examination data, and laboratory test results, a lower midline laparotomy was planned. Due to the unclear location of the gestational sac, the next steps of surgery were clarified intraoperatively.

Pelvic ultrasound showed the uterus to be 81 mm×59 mm×30 mm. The uterine cavity was up to 10 mm. On the right side of the uterus, a lesion was found measuring 104 mm×71 mm with a gestational sac measuring 62 mm, crown-rump length was 6.67 cm (week 13 of pregnancy). No free fluid was found in the pelvis. Ultrasound signs of ectopic (abdominal?) pregnancy were found.

Table 3 shows the results of complete blood counts performed in the Central District Hospital and the Emergency Department of the Kaluga Regional Clinical Hospital.

Table 3. Clinical blood test indicators of patient B

Indicators	Central Regional Hospital	Kaluga Regional Clinical Hospital
Erythrocytes, $\times 10^{12}$/l	2,3	2,15
Hemoglobin, g/l	75	66
Hematocrit, %	–	18,5
Platelets, $\times 10^9$/l	–	222
Leukocytes, $\times 10^9$/l	16,4	15,5

No abnormalities were found in the tests performed according to clinical guidelines and protocols (urinalysis, coagulation profile, blood biochemistry, blood serology).

When the abdominal cavity was opened, liquid blood and blood clots were found in the pelvis, abdominal cavity, flanks, and subhepatic space. The body of the uterus was round, up to six weeks of pregnancy in size. The left appendages included the fallopian tube and a structural ovary measuring 3.0 cm×2.5 cm×3.0 cm with no visible abnormalities. In the area of the right appendages, a neoplasm with a diameter of up to 16 cm was found. It contained a gestational sac with an embryo and chorionic tissues that were fixed and integrated into the ovary, with a tendency to be destroyed. The fallopian tube was difficult to locate because it was filled with clots. A right adnexectomy with suturing was performed. Surgical debridement was performed until a silicone drainage was placed in the recto-uterine pouch and drained through the right contraincision. The anterior wall was closed layer by layer. The skin was sutured using a Donati technique. During surgery, three doses of fresh frozen plasma and two doses of red cell mass were transfused as indicated.

Histology No. 33894-05 showed serous phlebitis in one of the umbilical veins (male fetus, length 8 cm, umbilical cord 4 cm), active chronic salpingo-oophoritis; chorionic villus invasion into the ovarian parenchyma with bleeding and necrosis; serous ovarian cysts.

The postoperative period was uneventful and the sutures were removed on day 10 with primary intention healing. The patient was discharged in satisfactory condition.

Figures 3 and 4 show images of this case.

Fig. 3. Right-sided ovarian pregnancy.

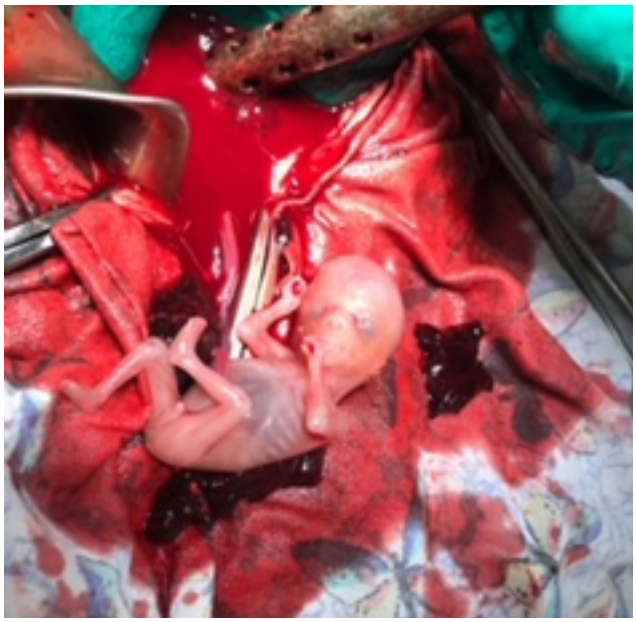

Fig. 4. 11–12-week embryo in ovarian pregnancy.

DISCUSSION

The gold standard of diagnostic criteria for ectopic pregnancy included presence of beta-HCG and trophoblasts found outside the uterus by pelvic ultrasound from day 18 of pregnancy. In practice, however, patients present with menstrual cycle disorders and a doubtful pregnancy test but cannot remember the date of their last period. If the period is delayed for three weeks, the absence of a gestational sac in the uterine cavity is a reason to reconsider the diagnosis and classify the patient as a high-risk group requiring follow-up, because the loss of time and underestimation of the condition severity can cost lives. This paper presents two clinical cases that clearly demonstrate the difficulty or lack of diagnosis in the preclinical stage of disease development. The first case was diagnosed exclusively during surgery, the second case was diagnosed after the appearance of clear clinical signs. These facts once again confirm the difficulty of diagnosing not only ectopic pregnancy, but especially its rare forms.

CONCLUSION

Two clinical cases of life-threatening ectopic pregnancy in rare locations are presented. Despite the modern, high-quality diagnostic tools available to women, the diagnosis was still challenging to establish and was ultimately made and confirmed only during surgery. It was therefore concluded that the clinical presentation may be the most significant factor influencing a healthcare provider's capacity to accurately diagnose and select an appropriate treatment strategy. Such rare sites, which are difficult to diagnose, should not be overlooked. If an ectopic pregnancy is not detected in a timely manner and the risks are underestimated, the outcome can be fatal.

Two case reports clearly demonstrate the importance of timely diagnosis before performing additional tests (pelvic ultrasound, beta-HCG) and appropriate choice of approach and extent of surgery.

The lack of awareness of ectopic pregnancy among women should also be mentioned. Many patients have no symptoms, do not remember the date of their last period, or do not monitor their menstrual cycle at all. Consequently, they often fail to seek medical attention in a timely manner. A combination of several factors can lead to a fatal outcome.

ADDITIONAL INFO

Authors' contribution. E.B. Ginzburg — surgical treatment of the patient, literature review, collection and analysis of literary sources, writing the text and editing the article, preparation of photographic material; B.G. Ginzburg — literature review, collection and analysis of literary sources, preparation and writing of the text of the article, editing the text of the article. All authors confirm that their authorship meets the international ICMJE criteria (all authors made a substantial contribution to the conception of the work, acquisition, analysis, interpretation of data for the work, drafting and revising the work, final approval of the version to be published and agree to be accountable for all aspects of the work).

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