

CASE REPORT

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Vanishing twin syndrome and chronic ruptured ectopic pregnancy in the setting of triplet heterotopic pregnancy

Quinn Rhodes, Wahibah Hannan, Jabez Gondokusumo, Sarah Hoopes

ABSTRACT

Introduction: The objective of this article is to present a case of a concurrent chronic ruptured ectopic pregnancy (CEP) and vanishing twin syndrome (VTS) in the setting of a naturally conceived heterotopic triplet pregnancy.

Case Report: A 31-year-old multiparous female presented with abdominal pain, vaginal bleeding, VTS, and an indeterminate mass in the posterior cul-de-sac. The diagnosis of a rare triplet heterotopic pregnancy (HP) was made during laparoscopy in the setting of VTS and CEP. The patient underwent laparoscopic left salpingectomy, and the remaining viable intrauterine pregnancy progressed to term vaginal delivery.

Conclusion: This case highlights the danger of relying on probability to rule out medical diagnoses and brevity of medical literature on CEP. Two intrauterine gestational sacs delayed arrival at the correct diagnosis of ectopic pregnancy due to the rarity of HP. In addition, providers did not recognize the classic signs of CEP: prolonged intermittent pelvic pain, vaginal bleeding, and an amorphous, avascular pelvic mass. Additional information regarding CEP should be included in common medical educational resources to better inform patient care.

Keywords: Chronic ruptured ectopic pregnancy, Heterotopic triplet pregnancy, Vanishing twin syndrome

How to cite this article

Rhodes Q, Hannan W, Gondokusumo J, Hoopes S. Vanishing twin syndrome and chronic ruptured ectopic pregnancy in the setting of triplet heterotopic pregnancy. J Case Rep Images Obstet Gynecol 2025;11(1):58–63.

Article ID: 100200Zo8QR2025

doi: 10.5348/100200Zo8QR2025CR

INTRODUCTION

A ruptured ectopic pregnancy is a gynecological emergency. This diagnosis is rarely missed in resource-rich settings. In this case report, we discuss the human fallibility of diagnosing a ruptured ectopic pregnancy in an atypical and unusual patient presentation. In this case, the patient presented with a chronic ruptured ectopic pregnancy and simultaneous vanishing twin syndrome (VTS) in the setting of a spontaneously conceived triplet heterotopic pregnancy (HP). All three of these conditions are uncommon diagnoses and the rarity of these conditions in combination presented a challenge for the diagnostician.

CASE REPORT

A 31-year-old Gravida 2 para 1 Hispanic female presented to the emergency department at her local hospital complaining of a 1-month history of left lower quadrant pain, vaginal bleeding, generalized abdominal cramping pain, nausea, and vomiting. Her last menstrual period was eight weeks prior to the presentation. This was the patient's first medical encounter since her positive home pregnancy test.

The patient reported regular menstrual cycles every 30 days with 5–7 days of bleeding and no hormonal contraception use within the last year. She denied a history of sexually transmitted infections or pelvic inflammatory disease. Her obstetric history included an uncomplicated

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Received: 06 January 2025

Accepted: 07 March 2025

Published: 17 April 2025

term spontaneous vaginal delivery. She denied the use of assisted reproductive technology (ART) to conceive this pregnancy. She had no significant medical history, surgical history, or social history.

On presentation, the patient was afebrile, normotensive, non-tachycardic, and appeared comfortable. Abdominal exam revealed tenderness to palpation of the lower quadrants bilaterally with no rebound or guarding. Pelvic exam revealed 5 mL of dark red blood in the vault with no active bleeding. A bimanual exam showed an anteverted 8-cm uterus, 1 cm cervical dilation, no adnexal tenderness, and no palpable masses.

On transvaginal ultrasound, the uterus measured 10.0 × 9.0 × 7.0 cm with two intrauterine gestational sacs. Sac A contained echogenic material with no fetal pole. Sac B showed a fetal pole with a crown-rump length measuring 22.58 mm corresponding to a gestational age of eight weeks and four days and a fetal heart rate of 167 bpm. A lambda sign, characteristic of a dichorionic diamniotic twin pregnancy, was observed between the gestational sacs. Neither ovary was identified on the ultrasound. A solid-appearing isoechoic focus, with no internal vascularity and measuring 5.8 × 4.8 × 5.0 cm, was observed posterior to the uterus (Figure 1). Free fluid was not noted. The patient's laboratory values included a hemoglobin of 10.5 g/dL, hematocrit of 31.4%, white blood cell count of 11,500 μ L, and beta-human chorionic gonadotropin (β -hCG) of 93,303.60 mIU/mL.

Given concern for possible ovarian torsion, laparoscopy was performed. Heterotopic pregnancy was not considered for the preoperative differential diagnosis. On entry into the abdominal cavity, 200 mL of clotted fibrinous blood was noted in the pelvis, surrounded by adhesions between the left fallopian tube, left ovary, posterior uterus, and anterior rectum. The left fallopian tube was dilated and ruptured with trophoblastic appearing tissue protruding through the wall positioned posterior to the uterus (Figure 2). The left ovary contained a cyst consistent with a corpus luteum. Slow oozing was noted from the left fimbriae. The right fallopian tube and ovary appeared normal. Intraoperative findings

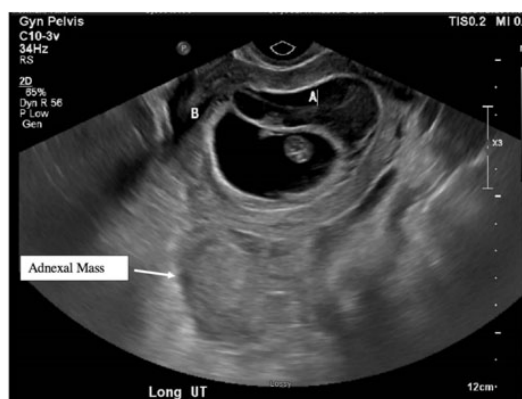


Figure 1: Additional longitudinal view of the uterus on transvaginal ultrasound showing two intrauterine gestational sacs and an indeterminate heterogenous, adnexal mass posterior to the uterus.

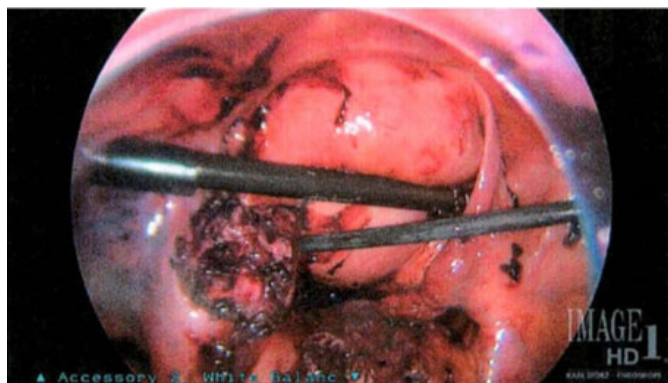


Figure 2: Laparoscopic image from examination of left fallopian tube and ovary with trophoblastic tissue adherent to the right adnexa.

were consistent with ruptured ectopic pregnancy and the decision was made to proceed with definitive treatment with left salpingectomy. After the case's conclusion, the fetal heart rate of sac B was confirmed to be 142 bpm. The left fallopian tube was sent to pathology, confirming ectopic pregnancy.

The patient's postoperative course was uncomplicated. She was discharged home independently the same day. At 2-week postoperative follow-up, the patient was recovering well. Ultrasound revealed a single gestational sac with cardiac activity. The remainder of the patient's pregnancy was uncomplicated. She had a term spontaneous vaginal delivery at 38 weeks with the delivery of a healthy male infant weighing 3380 grams. After delivery, the patient provided written consent to publish all medically pertinent information.

DISCUSSION

This case presents an intersection of two uncommon diagnoses, chronic ectopic pregnancy, and VTS, with an exceedingly rare diagnosis, spontaneous heterotopic triplet pregnancy. The rarity of these conditions in combination presents a challenge for the diagnostician. We will discuss separately the incidence, clinical presentation, and diagnostic challenges of these conditions.

The incidence of twin HP is 1/30,000 among patients who conceived spontaneously and 1.5/1000 among patients who used assisted reproductive technology [1]. Due to the rarity of triplet HP, there are no data on its incidence. At the time of publication of this article, there have been 11 documented cases in the literature of spontaneously conceived triplet HP pregnancy [2–12] (Table 1). The majority of these cases presented in the first trimester, while only two occurred in the second trimester at 15 and 18 weeks. Our case is the only documented triplet HP case that presented with acute abdominal pain, overt vaginal bleeding, and VTS at the time of initial presentation. Of note, the case report published by Arsala et al. describes a triplet HP case with VTS. However, the

Table 1: Confirmed spontaneous triplet heterotopic pregnancies (POD = postoperative day)

Primary author	Date	Country	Clinical presentation	Ultrasound findings	Gestational age at presentation	Location of pregnancies	Treatment	Outcome of intrauterine pregnancy(s)	Ruptured ectopic pregnancy
Alsunaidi	2005	Saudi Arabia	Lower abdominal pain for eleven days	Gestational sac in left adnexa with two yolk sacs	8 weeks	Single intrauterine pregnancy, twin left tubal pregnancies	Laparotomy, left salpingectomy	Term vaginal delivery of viable neonate at 39 weeks	Yes
Cholkeri-Singh	2007	USA	Acute onset right lower abdominal pain	No adnexal mass seen; free fluid in the right colonic gutter	5 weeks	Twin diamniotic dichorionic intrauterine pregnancies. Single right tubal pregnancy.	Laparoscopic right salpingectomy	Primary cesarean section of two viable infants at 34 weeks due to pre-term labor with fetal malpresentation	Yes
Simsek	2008	Turkey	Lower abdominal pain, positive for peritoneal signs	Free fluid in posterior cul de sac	9 weeks	Twin mono-amniotic mono-chorionic right tubal pregnancies, single intra-uterine pregnancy.	Exploratory laparotomy with right salpingectomy	Spontaneous term vaginal delivery of viable infant	Yes
Arsala	2014	Australia	Diffuse abdominal pain, distended abdomen, rebound tenderness and guarding on exam.	Two intrauterine gestational sacs and one right adnexal gestational sac, all three with fetal heart rate. Free fluid in posterior cul-de-sac.	6 weeks	Twin intrauterine pregnancies, single right tubal pregnancy	Laparoscopic right salpingectomy	Vanishing twin syndrome of twin B diagnosed on POD8, healthy ongoing pregnancy of twin A	Yes
Nnoli	2015	USA	Initially asymptomatic, diagnosed by ultrasound at clinic visit. The patient was scheduled for surgery for next day, overnight patient developed acute abdominal with peritoneal signs on exam.	Outpatient ultrasound: ectopic pregnancy in right fallopian tube, near proximity to the right ovary. No repeat ultrasound performed due to high clinical suspicion of ruptured ectopic pregnancy.	8 weeks	Twin intrauterine pregnancies, single right distal tubal pregnancy with involvement of broad ligament	Laparoscopic right fimbriectomy	Ongoing twin dichorionic diamniotic intra-uterine pregnancy at 24 weeks	Yes
Rengaraj	2016	India	Vaginal spotting, mild abdominal pain on abdominal exam, no peritoneal signs	Complex mass right adnexal mass, minimal free fluid	8 weeks	Twin intrauterine pregnancy, single right tubal ectopic pregnancy	Exploratory laparotomy, right salpingectomy	Term vaginal delivery of viable twins at 37 weeks	No
Kotlyar	2016	USA	Abdominal pain for four days	Heterogenous right adnexal mass, minimal free fluid on ultrasound	15 weeks	Twin monochorionic-monoamniotic Intrauterine pregnancy. Single right tubal pregnancy	Laparoscopic right salpingectomy	Fetal demise of both twins at 22 weeks	No

Table 1: (Continued)

Primary author	Date	Country	Clinical presentation	Ultrasound findings	Gestational age at presentation	Location of pregnancies	Treatment	Outcome of intrauterine pregnancy(s)	Ruptured ectopic pregnancy
Maheshgir	2017	India	New onset right lower quadrant abdominal pain	Heterogenous right adnexal mass 6.8 × 4.6 cm, free fluid noted on ultrasound	10 weeks	Twin dichorionic-diamniotic intrauterine pregnancy, demise of twin A. Single right tubal pregnancy.	Laparoscopic right salpingectomy	No comment on outcome of remaining viable intrauterine pregnancy	Yes
Torky	2017	Egypt	Abdominal pain	Right adnexal mass 4 × 8 cm	10 weeks	Twin intra-uterine pregnancy. Single right tubal pregnancy.	Laparoscopic right salpingostomy	Vaginal pre-term delivery of two viable infants at 34 weeks	Yes
Guimarães	2019	Brazil	Acute right lower abdominal pain, rebound tenderness	Tubular structure 2cm in size in right iliac fossa	8 weeks	Twin dichorionic-diamniotic intrauterine pregnancy. Single right tubal pregnancy.	Exploratory laparotomy, right salpingectomy	Primary cesarean section of two viable infants at 36 weeks	Yes
Nkurunziza	2020	Rwanda	Diffuse abdominal pain, hypovolemic shock	No cardiac activity of all three fetuses.	18 weeks	Twin monochorionic-diamniotic intrauterine pregnancy. Single right sided interstitial pregnancy.	Exploratory laparotomy, subtotal hysterectomy due to extent of cornual rupture from interstitial pregnancy	No viable pregnancy remaining after subtotal hysterectomy	Yes

VTS was diagnosed eight days after surgical treatment and was not present at the time of initial presentation with triplet HP.

Among ectopic pregnancies, 20% are classified as chronic. Chronic ectopic pregnancy (CEP) is an ectopic pregnancy with a growth pattern characterized by a protracted cycle of microvascular trauma, bleeding, and thrombus formation that culminates in an enlarging pelvic hemocoel. The theorized mechanism of chronicity is that the trophoblastic invasion impacts terminal vascular branches, which results in slow bleeding that can be outpaced by the coagulation cascade [3]. The chronicity of the condition can cause an atypical presentation with poorly localized, intermittent, dull pain. This presentation contrasts sharply with the classic presentation of acute ruptured ectopic pregnancy which presents with constant, sharp adnexal pain making diagnosis difficult. Further confounding the diagnosis, CEP can present with undetectable serum β-hCG and usually will not present with free fluid on pelvic ultrasound, which can be falsely reassuring [13]. The diagnosis is usually made during evaluation of the amorphous adnexal mass via diagnostic laparoscopy [14]. Of note, information on CEP is sparse in mainstream didactic literature. Neither the ACOG Practice Bulletin, PubMed StatPearls, nor UpToDate articles on ectopic pregnancy mention the variant presentation of CEP. Williams Gynecology textbook only has two paragraphs dedicated to the topic of CEP [14]. Consequently, many providers may be unfamiliar with

CEP and unable to associate a similar clinical presentation with a chronic ectopic pregnancy.

Vanishing twin syndrome occurs in approximately one-third of twin pregnancies and half of triplet or higher-order pregnancies [15]. It is defined as the occurrence of the spontaneous reduction of one or more embryos in a multi-gestation pregnancy, resulting in a reduced number of viable embryos. The majority of epidemiological data on VTS is based on ART-conceived pregnancies, as those pregnancies are more likely to be followed by serial ultrasounds. The clinical presentation of VTS varies from asymptomatic to overt symptoms of vaginal bleeding, cramping, and pelvic pain with a correlating span of physical exam findings from a closed cervix with no bleeding to a dilated and effaced cervix [15]. The proportion of asymptomatic VTS cases to symptomatic VTS cases has not been addressed in the existing literature. Even with overt physical manifestations, the remaining viable pregnancy or pregnancies can be carried to full-term gestation. In this case, the patient’s clinical symptoms, physical exam, and intrauterine ultrasound findings aligned with the diagnosis of VTS. The adnexal mass could not be attributed to VTS, and thus prompted further investigation.

CONCLUSION

This case presents a diagnostic challenge for several reasons: the rarity of the final diagnosis of triplet

HP, chronic ectopic pregnancy presentation, and the distracting presence of a VTS. Fortunately, the presence of a poorly defined pelvic mass, found to be the pelvic hematocele from a CEP, prompted further investigation that culminated in the appropriate treatment. Several key lessons should be gained from this case. First, the astute clinician must understand that statistical improbability does not equate to impossibility. This principle is most salient for medical conditions such as ectopic pregnancy, which when unrecognized can result in fatality. Thus, the statistical improbability of a heterotopic triplet pregnancy should not definitively rule out the diagnosis of ectopic pregnancy. Second, this case unveiled a knowledge gap in our current gynecological graduate medical education system: CEP.

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Author Contributions

Quinn Rhodes – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Wahibah Hannan – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Jabez Gondokusumo – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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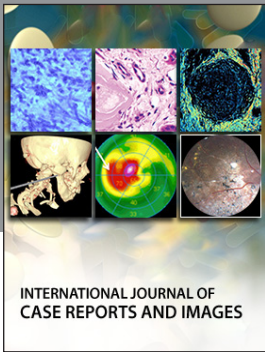
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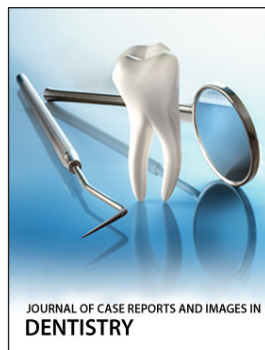
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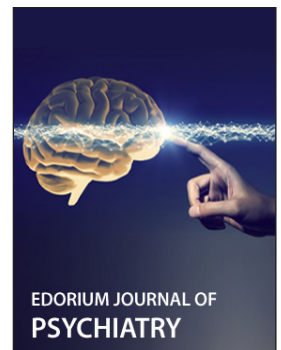
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