

Unilateral tubal twin ectopic pregnancy: a case report and literature review

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Received: 19 Aug 2026, **Accepted:** 05 Sep 2026, **Published:** 09 Sep 2026

Citation: Gaye Yaye Fatou Oumar, N. Niang, N. S. Diop, et al. Unilateral tubal twin ectopic pregnancy: a case report and literature review. *Obs Gyn Cur Res.* 2026; 1(1), 01-05.

Abstract

Twin ectopic pregnancy is a rare entity, and its unilateral tubal presentation remains exceptional. We report the case of a 30-year-old woman, gravida 2 para 1, admitted for pelvic pain with 6 weeks of amenorrhea. Ultrasound revealed an unruptured evolving twin ectopic pregnancy at 7 weeks and 1 day of gestation, with a plasma β -hCG level of 42,750 IU/L. Laparoscopy revealed a fissured right-sided ectopic pregnancy managed by total salpingectomy, and histopathological examination confirmed the twin nature of the pregnancy. Through this case and a review of the literature, we discuss the diagnostic and therapeutic particularities of this rare condition.

Keywords: ectopic pregnancy, twin pregnancy, unilateral, tubal, β -hCG, laparoscopy

Introduction

Ectopic pregnancy (EP) is defined as the implantation of a fertilized ovum outside the endometrial cavity. It complicates approximately 1 to 2% of all pregnancies and represents one of the leading causes of maternal death during the first trimester, while significantly compromising subsequent fertility [1, 2]. The fallopian tube is the most common implantation site, accounting for more than 90% of cases [3].

While single-fetus ectopic pregnancy is a well-known condition, its twin form is a much rarer entity, first described in 1891 by De Ott [4]. Since then, only about a hundred cases of twin ectopic pregnancies have been reported in the medical literature, most of which were diagnosed intraoperatively or postoperatively [5, 6, 7, 8]. The incidence of unilateral twin ectopic pregnancy ranges from 1/20,000 to 1/125,000 pregnancies, corresponding to a rate of approximately 1 case per 725 to 1,580 tubal pregnancies [1, 3, 4, 5, 9]. In recent decades, the prevalence of ectopic pregnancy has increased owing to the rise in sexually transmitted infections and pelvic surgeries, and above all to the use of assisted reproductive technology [1, 5, 6]. However, in nearly half of reported cases, no identifiable risk factor is found [3]. The exact pathogenesis of spontaneous unilateral ectopic pregnancies is not yet fully

understood. Two main mechanisms have been proposed: monozygotic twinning occurring after implantation of the fertilized egg in the tube, or the synchronous implantation of two distinct fertilized ova in the same tube [5]. The “great mimic of the abdomen,” as Mondor called it, is even more difficult to diagnose in its twin form, which calls for heightened vigilance. Unilateral twin ectopic pregnancy carries a higher risk of maternal morbidity and mortality, which justifies early diagnosis through the combination of ultrasound and quantitative human chorionic gonadotropin (hCG) assay, as well as prompt intervention to prevent potentially life-threatening complications, particularly tubal rupture [3].

We report a new case of unilateral tubal twin ectopic pregnancy to contribute to the epidemiological data on this exceptional condition. We will discuss the clinical, paraclinical, and therapeutic particularities of this entity in light of the literature.

Case Report

Mrs. S. L., a 30-year-old woman, gravida 2 para 2, was seen in the gynecological emergency department for acute pelvic pain occurring in the context of 6 weeks of secondary

amenorrhea based on the date of her last menstrual period. History-taking revealed no notable medical or surgical history, nor any history of contraceptive use. She was not exposed to tobacco.

Physical examination found a hemodynamically stable patient. The abdomen showed no scar from previous surgery; palpation revealed guarding and tenderness in the right iliac fossa and periumbilical tenderness. Gynecological examination revealed tenderness on palpation of the pouch of Douglas.

Laboratory workup showed a plasma β -hCG level of 42,750 IU/L and a hemoglobin level of 11.7 g/dL. Transvaginal pelvic ultrasound showed an empty uterus and a right latero-

uterine image suggestive of a gestational sac containing two embryos with cardiac activity, along with a trophoblastic mass without a visible inter-amniotic membrane. There was no associated hemoperitoneum.

Given this presentation combining clinical signs of peritoneal irritation, a marked elevation of β -hCG, and the ultrasound findings, we concluded that this was an evolving, fissured twin ectopic pregnancy at 7 weeks of amenorrhea (WA) + 1 day. Surgical management by laparoscopy was decided upon. Intraoperative exploration confirmed a fissured right-sided ectopic pregnancy. A total right salpingectomy was performed. The postoperative course was uneventful. Histopathological examination of the surgical specimen confirmed the diagnosis of twin ectopic pregnancy.



Figure 1: Ultrasound image Evolving twin ectopic pregnancy

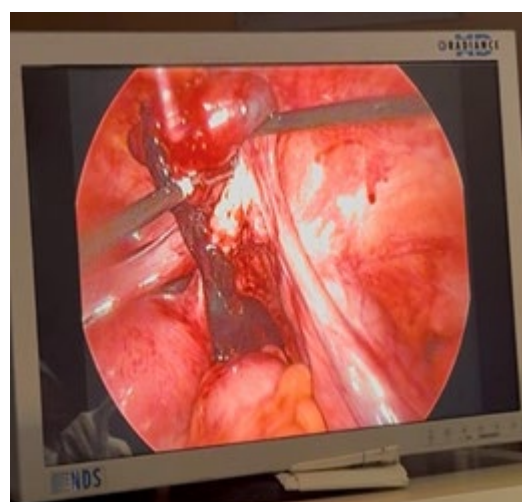


Figure 2: Intraoperative laparoscopic image Fissured right ampullary ectopic pregnancy

Discussion

Unilateral twin ectopic pregnancy is a particularly rare condition, first described in 1891 by the Russian obstetrician-gynecologist Ott [6]. Since then, only about a hundred cases have been published in the world medical literature [6,7]. The exact prevalence of unilateral ectopic twin pregnancy remains poorly known, as some cases may go unnoticed in the absence of early ultrasound diagnosis prior to any management. In most cases, the diagnosis of unilateral twin ectopic pregnancy is made intraoperatively or postoperatively [8]. Since the advent of medical treatment of ectopic pregnancy with methotrexate, many cases of ectopic pregnancy no longer undergo histopathological examination [9]. Such examination would have confirmed the diagnosis of ectopic pregnancy and determined the nature of the pregnancy (singleton, twin, or multiple; molar or not). The spontaneous nature of this form of ectopic pregnancy makes it exceptional. The prevalence of spontaneous unilateral twin ectopic pregnancy is estimated at between 1/125,000 and 1/250,000 spontaneous pregnancies [1,6,9]. According to reported cases, embryonic cardiac activity may be present or absent in one or both twins [3]. Unilateral ectopic twin pregnancy with two live embryos remains anecdotal. In 2022,

only eight such cases had been reported in the literature [6]. The systematic review by Sidhar et al. [5] in 2025 identified seventeen published clinical cases of unilateral ectopic pregnancies with live twins from spontaneous conception, collected from several countries (United Kingdom, United States, India, Iran, South Korea, and Ethiopia). Our observation falls within this exceptional group. To our knowledge, this is the first reported case of an evolving unilateral twin ectopic pregnancy in Senegal and in West Africa. Gure et al. [3] reported the first African case, which occurred in Ethiopia in 2022, followed by Mounguengui et al. [2] in Gabon in 2025.

The risk factors classically implicated in the occurrence of ectopic pregnancy include a history of pelvic inflammatory disease, tubal or pelvic surgery, sexually transmitted infections—particularly Chlamydia trachomatis infection—smoking, assisted reproductive technology, and intrauterine device use [1, 5, 6]. However, in nearly half of reported cases, no identifiable risk factor is found [3]. This was the case for our patient, who had no risk factors for ectopic pregnancy. The main risk factor for ectopic pregnancy in Africa remains sexually transmitted infections, as shown by the recent

publication of Mounquengui et al. [2] in Gabon.

Ectopic pregnancy remains the leading gynecological emergency of the first trimester of pregnancy, affecting 1.3 to 2.4% of pregnancies [1, 8]. Its clinical presentation is variable, ranging from a complete absence of symptoms to more pronounced unilateral pelvic discomfort, or even tubal rupture leading to hemorrhagic shock [8]. It must therefore be recognized and managed early. Its twin form is all the more difficult to diagnose as it is dangerous. Indeed, because of the twin pregnancy, the size of the products of conception is greater than would be expected for the gestational age. In addition, the limited distensibility of the tube considerably increases the risk of tubal rupture [5]. Unilateral twin ectopic pregnancy carries a markedly higher risk of rupture, observed in approximately 30 to 50% of cases, hence the importance of very early diagnosis [7]. In the review by Sidhar et al. [5], gestational age at diagnosis ranged from six to twelve weeks and six days of amenorrhea. Diagnosis was made very early in our patient, at 6 weeks of amenorrhea, as in the cases of Salloum et al. [7] in Russia, Betti et al. [9] in Italy, Lategan et al. [6] in New Orleans, and Li et al. [4] in Canada. In contrast, in the case reported by Mounquengui et al. [2] in Gabon, diagnosis was made at a more advanced gestational age, at 9 weeks of amenorrhea. Diagnosing ectopic pregnancy can prove very difficult, especially at an ultra-early gestational age (6 weeks). The combination of plasma β -hCG assay and pelvic ultrasound remains indispensable whenever ectopic pregnancy is suspected [1, 2]. In our case, the plasma β -hCG level was particularly high (42,750 IU/L), corroborating the level reported by Mikou et al. [10] in Morocco (42,090 IU/L). Suspicion of an extrauterine implantation increases when transvaginal ultrasound fails to demonstrate an intrauterine gestational sac despite a β -hCG concentration equal to or above the usual discriminatory threshold (approximately 1,500 IU/L) [4,10]. As a general rule, β -hCG levels relative to gestational age observed in ectopic pregnancies are lower than those of normal intrauterine pregnancies; however, twin forms may be accompanied by disproportionately high levels, mimicking those of an early intrauterine pregnancy and thereby delaying diagnosis [4,10]. This phenomenon, reported by several authors with initial β -hCG levels ranging from 1,750 to over 127,000 IU/L depending on the series, constitutes a classic diagnostic pitfall, as illustrated by the case reported by Mikou et al. [10], in whom an initially normal ultrasound combined with an atypical rise in β -hCG required close monitoring before two extrauterine gestational sacs were subsequently identified [5, 10]. In our observation, given an empty uterus on transvaginal ultrasound and despite a β -hCG value markedly elevated for an ectopic pregnancy (42,750 IU/L), suspicion of a multiple ectopic pregnancy was warranted. Practitioners should therefore consider a twin form of ectopic pregnancy as a first-line differential diagnosis when β -hCG values are abnormally elevated [5]. However, this rule is not absolute. Some authors have encountered diagnostic difficulties. In the case reported by Rolle et al. [11], plasma β -hCG was 263 mIU/mL, and it was transvaginal ultrasound that established the diagnosis, suggesting a nonviable unilateral twin ectopic pregnancy.

Thus, ultrasound, particularly transvaginal ultrasound, is a valuable tool that, in expert hands, allows the diagnosis of ectopic pregnancy to be made. It remains the first-line investigation, with reported sensitivity between 87 and 99% and specificity between 94 and 99.9% for the diagnosis of unilateral twin tubal ectopic pregnancy [5, 9]. The most characteristic direct sign is the demonstration, within a heterogeneous or solid-cystic adnexal mass, of two thick-walled sacular images corresponding to the gestational sacs, or of a single gestational sac containing two embryos [9]. Detection of a fetal heart rate provides additional, non-negligible confirmatory evidence [1, 7]. Color Doppler, by revealing a “ring of fire” pattern reflecting peritrophoblastic hypervascularization, also helps distinguish an evolving ectopic pregnancy from a hemorrhagic corpus luteum [5]. Despite these advances, achieved through improvements in ultrasound machine resolution in recent years, precise preoperative diagnosis remains rare, with fewer than 10% of cases described in the literature having been identified before surgery [5, 9].

In our case, a single gestational sac containing two yolk sacs and two embryonic echoes with cardiac activity was observed, consistent with a monochorionic twin pregnancy. Although the number of yolk sacs is sometimes used to determine amnionicity in early pregnancy, this criterion is not always reliable, and the absence of a visible amniotic membrane at this stage does not exclude diamnionicity, as ultrasound identification of this membrane can be delayed [3]. Consequently, although monochorionicity was confirmed in our observation, amnionicity could not be definitively determined.

Topographically, a clear predominance of right tubal involvement has been reported by Ophir et al. [12], Sousa et al. [13], and Sidhar et al. [5], consistent with our observation. This predominance may reflect the physiological dominance of right-sided ovulation or anatomical differences in tubal vascularization; furthermore, greater blood pooling in the left ovary, related to anatomical particularities, could raise local ovarian temperature and reduce the fertilizing potential of oocytes on that side, which could help explain the right-sided predominance observed in the literature [5]. Unilateral tubal twin pregnancies carry a markedly higher risk of tubal rupture than singleton ectopic pregnancies, estimated at between 30 and 50% of cases depending on the series, owing to the increased distension of the tube by the two gestational sacs and the tube's limited capacity for expansion [3, 5]. The classic triad of amenorrhea, pelvic pain, and metrorrhagia is found in fewer than half of cases [2, 9]. In our observation, the patient presented with pelvic pain and clinical signs of peritoneal irritation (periumbilical tenderness, tenderness of the pouch of Douglas), consistent with the intraoperative finding of a fissured ectopic pregnancy, despite a preoperative ultrasound that had concluded to an unruptured form. This discrepancy highlights the possible rapid progression of these pregnancies, which carry a high risk of rupture, and the need for prompt surgical management as soon as the diagnosis is suspected.

Management strategies depend on the patient's hemodynamic stability, serum β -hCG levels, as well as the size and location of the extrauterine mass and the progression of the pregnancy [7]. Three therapeutic options are classically described for the management of ectopic pregnancy: expectant management with monitoring, medical treatment with methotrexate, and surgical treatment [1]. In twin forms, the therapeutic choice remains debated in the absence of specific recommendations, as the rarity of the condition precludes large-scale randomized studies [4, 5].

Medical treatment with methotrexate has been successfully proposed in selected cases of stable patients without embryonic cardiac activity and with moderate initial β -hCG levels, whether using a single-dose or multiple-dose protocol [4, 5, 9]. However, several failures of this treatment have been reported, particularly in the presence of embryonic cardiac activity or high initial β -hCG levels (above 5,000–13,000 IU/L), resulting in secondary tubal rupture requiring emergency surgery [5, 9]. The progression of the pregnancy therefore constitutes a strong relative contraindication to medical treatment, particularly in twin forms where the risk of rupture is increased [5, 13]. The case of Betti et al. [9] illustrates this perfectly; despite repeated methotrexate injections (3 doses), the pregnancy continued to progress, ultimately resulting in tubal rupture requiring salpingectomy. These findings underscore the need for caution when using intramuscular methotrexate in cases of twin ectopic pregnancy, weighing the goal of fertility preservation against the increased risk of rupture and the possibility of treatment failure [5]. Because of the low incidence of spontaneous unilateral tubal twin pregnancies, no consensus has been established regarding the best management option [6]. For example, some authors have reported the use of uterine artery embolization after failure of medical treatment with methotrexate, particularly in interstitial forms [12]. In contrast, Li et al. [4] reported a case of twin ectopic pregnancy with simultaneous interstitial and tubal implantation in a hemodynamically stable patient, successfully treated with multiple-dose systemic methotrexate.

Recent data also confirm that a high β -hCG level, the presence of a multiple pregnancy, and a large extrauterine mass are associated with a lower success rate of methotrexate treatment, which justifies surgical management in these situations [7].

Surgical treatment remains the standard of care in most published series, particularly in cases of hemodynamic instability, confirmed tubal rupture, or high β -hCG levels [5, 7]. Laparoscopy is preferred over laparotomy in hemodynamically stable patients because of its better cost-effectiveness, lower operative morbidity, shorter operating and hospitalization times, and faster recovery [1, 7]. However, in developing countries, laparoscopic surgery—the reference treatment for ectopic pregnancy remains limited in practice due to lack of available equipment or delayed presentation for care. Mounguengui et al. [2] had to resort to

laparotomy to manage their case of fissured unilateral twin ectopic pregnancy.

Total salpingectomy remains the procedure most frequently performed, particularly in cases of tubal rupture or associated hemoperitoneum, whereas conservative salpingostomy may be considered in patients wishing to preserve their fertility, in the absence of contraindications [1, 6, 7]. The markedly higher risk of rupture in twin ectopic pregnancies, estimated at 30 to 50% of cases, reinforces the predominant role of surgery in the therapeutic arsenal [3, 5]. In our patient, the intraoperative finding of a fissured tube justified performing a total salpingectomy, the procedure best suited to this situation to ensure satisfactory hemostasis and prevent the risk of persistent trophoblastic tissue. As data supporting salpingostomy in twin ectopic pregnancies remain limited, the current literature favors salpingectomy as a safer and more definitive approach [7]. Nevertheless, salpingostomy remains strongly recommended in the presence of infertility risk factors [1].

Data from the literature suggest that maternal prognosis is generally favorable when diagnosis and management are early, with no maternal deaths reported in the main recent series [5]. Regarding subsequent fertility, data comparing salpingectomy and salpingostomy specifically in the context of twin ectopic pregnancies remain limited, but studies on singleton ectopic pregnancies have found no significant difference in subsequent intrauterine pregnancy rates between the two surgical approaches [1, 5].

Conclusion

The occurrence of a spontaneous unilateral ectopic pregnancy with viable twins is a rare and perilous event that poses significant diagnostic and therapeutic challenges in modern obstetrics. While singleton ectopic pregnancy is already considered an extreme emergency because of the risk of hemorrhage, unilateral twinning increases this danger further, exposing the patient to earlier and more severe tubal rupture and to increased maternal morbidity and mortality. Although limited, the data in the literature outline a recognizable clinical profile: an early gestational age at diagnosis, disproportionately elevated β -hCG levels, a predominance of right tubal involvement, and frequent recourse to surgery.

The real challenge for the gynecologist, beyond always considering the possibility of ectopic pregnancy, is to diagnose the unilateral twin form early, using the combination of transvaginal ultrasound and quantitative β -hCG assay. Individualized management, ideally laparoscopic, must ensure that preservation of life takes priority over preservation of fertility for these patients.

Better knowledge of this condition and the publication of new cases will help improve future diagnostic and therapeutic strategies.

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