

# Unilateral live twin tubal ectopic pregnancy presenting at 12 weeks of gestation

## A case report

Chen-June Seak, MD<sup>a,b,\*</sup>, Zhong Ning Leonard Goh, MBBS<sup>c</sup>, Alexis Ching Wong, MBBS<sup>c</sup>, Joanna Chen-Yeen Seak, MD<sup>c</sup>, Chen-Ken Seak, MBBS<sup>c</sup>

### Abstract

**Rationale:** Abdominal pain in pregnancy represents a demanding diagnostic challenge in the emergency department (ED) due to the extensive list of differential diagnoses to be considered, coupled with the possibility of each disease having nonclassical, atypical signs and symptoms, resultant from the patient's pregnant state. Additionally, emergency physicians (EPs) face limitations on investigative imaging modalities because of the need to minimize fetal radiation exposure. EPs have to tackle this diagnostic challenge while performing a balancing act to maximize both maternal and fetal outcomes in a time-sensitive manner, because any delays in decision-making at the ED may threaten the safety of mother and child. Two common causes of abdominal pain in pregnancy presenting to the ED are acute appendicitis and ectopic pregnancy. The latter is almost always diagnosed by 10 weeks of gestation. Here, we report an extremely rare case of unilateral live spontaneous twin tubal ectopic pregnancy presenting past 12 weeks of gestation, diagnosed after magnetic resonance imaging (MRI) of the abdomen.

**Patient concerns:** A 37-year-old gravida 2 para 1 at 12 weeks and 6 days of gestation presented to our ED with a 2-day history of right iliac fossa pain, not associated with vaginal bleeding, fever, diarrhea, and vomiting. On examination, she was tachycardic (pulse rate 124 beats/min) and hypertensive (blood pressure 142/88 mm Hg). There was marked tenderness and guarding at the lower abdomen.

**Diagnoses:** Blood investigations were unremarkable, while abdominal ultrasonography found a live twin gestation with foetal heartbeats of 185 and 180 beats/min. MRI of the abdomen revealed an empty uterine cavity; 2 amniotic sacs and fetuses of diameter 10 cm, and a single placenta were noted in the right uterine adnexa. The patient was diagnosed with right live monochorionic diamniotic twin tubal pregnancy.

**Intervention:** Our patient underwent emergency laparoscopic right salpingectomy.

**Outcomes:** The operation was successful and her postoperative care remained uneventful up to discharge.

**Lessons:** Ectopic pregnancy cannot be ruled out based on prior normal antenatal examinations and gestational age of >10 weeks. EPs should not hesitate to order MRI scans for further evaluation if ultrasonography and laboratory findings are equivocal.

**Abbreviations:** ED = emergency department, EP = emergency physician, MRI = magnetic resonance imaging.

**Keywords:** diagnostic pitfall, ectopic pregnancy, emergency department, live twin tubal pregnancy

Editor: N/A.

The authors have no conflicts of interest to disclose.

<sup>a</sup> Department of Emergency Medicine, Lin-Kou Medical Center, Chang Gung Memorial Hospital, <sup>b</sup> College of Medicine, Chang Gung University, Taoyuan, Taiwan, Republic of China, <sup>c</sup> Sarawak General Hospital, Kuching, Sarawak, Malaysia.

\* Correspondence: Chen-June Seak, Department of Emergency Medicine, Lin-Kou Medical Center, Chang Gung Memorial Hospital, No. 5, Fusing St., Guei-shan Township, Taoyuan County 333, Taiwan, Republic of China (e-mail: julianseak@hotmail.com).

Copyright © 2019 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and buildup the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

How to cite this article: Seak CJ, Goh ZNL, Wong AC, Seak JCY, Seak CK. Unilateral live twin tubal ectopic pregnancy presenting at 12 weeks of gestation. *Medicine* 2019;98:38(e17229).

Received: 31 January 2019 / Received in final form: 28 July 2019 / Accepted: 26 August 2019

<http://dx.doi.org/10.1097/MD.0000000000017229>

## 1. Introduction

Abdominal pain in pregnancy represents a demanding diagnostic challenge in the emergency department (ED). This is due to the extensive list of differential diagnoses—encompassing medical, surgical, gynecological, obstetrical, and psychiatric diseases—to be considered, coupled with the possibility of each disease having nonclassical, atypical signs and symptoms, resultant from the patient's pregnant state. Additionally, emergency physicians (EPs) face limitations to investigative imaging modalities because of the need to minimize fetal radiation exposure.<sup>[1]</sup> EPs have to tackle this diagnostic challenge while performing a balancing act to maximize both maternal and fetal outcomes in a time-sensitive manner, because any delays in decision-making at the ED may threaten the safety of mother and child.<sup>[2]</sup>

Acute appendicitis is the most common surgical disease encountered during pregnancy,<sup>[3]</sup> whereas ectopic pregnancy has an incidence of up to 16% in the ED.<sup>[4]</sup> The similarities in clinical presentation between both diseases often result in diagnostic dilemmas for EPs managing pregnant ladies presenting to the ED with right lower abdominal pain early in the first

trimester. Distinction between these 2 disorders becomes easier as the period of gestation gets longer, because almost all ectopic pregnancies are diagnosed by 10 weeks of gestation.<sup>[5]</sup> Here, we report an extremely rare case of unilateral live spontaneous twin tubal ectopic pregnancy presenting past 12 weeks of gestation. This case report was approved by the Chang Gung Medical Foundation Institutional Review Board (IRB 201800934B0). Informed consent was obtained from the patient for publication of this case report and accompanying images.

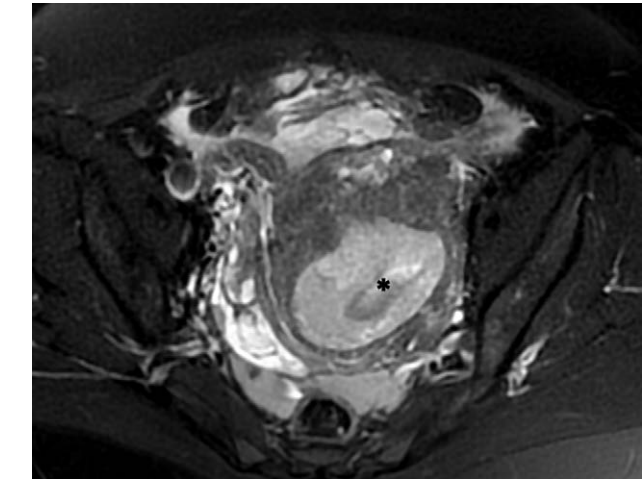
## 2. Case presentation

A 37-year-old gravida 2 para 1 at 12 weeks and 6 days of gestation presented to our ED with a 2-day history of right iliac fossa pain, not associated with vaginal bleeding, fever, diarrhea, and vomiting. Her previous antenatal examinations at other healthcare facilities were reported to be normal. There was no history of infertility treatment, and also risk factors of ectopic pregnancy in her surgical and gynecologic history. On examination, she was tachycardic (pulse rate 124 beats/min) and hypertensive (blood pressure 142/88 mm Hg). There was marked tenderness and guarding at the lower abdomen. Laboratory results were unremarkable. Transabdominal ultrasound at current presentation revealed a live twin gestation with detectable fetal cardiac activity of 185 and 180 beats/min, respectively. Each crown-rump length corresponded to 12 weeks 4 days and 12 weeks 1 day (Fig. 1, arrowheads), respectively.

Acute appendicitis was considered but ultimately ruled out after magnetic resonance imaging (MRI) of the abdomen revealed an empty uterine cavity (Fig. 2, asterisk). Two amniotic sacs and fetuses of diameter 10cm (Fig. 3, arrows) and a single placenta (Fig. 3, arrowhead) were noted in the right uterine adnexa. The patient was diagnosed with right live monochorionic diamniotic twin tubal pregnancy and underwent emergency laparoscopic right salpingectomy. Her postoperative care remained uneventful up to discharge.

## 3. Discussion

Ectopic pregnancy refers to the implantation of a developing blastocyst outside the uterine endometrial lining, most frequently

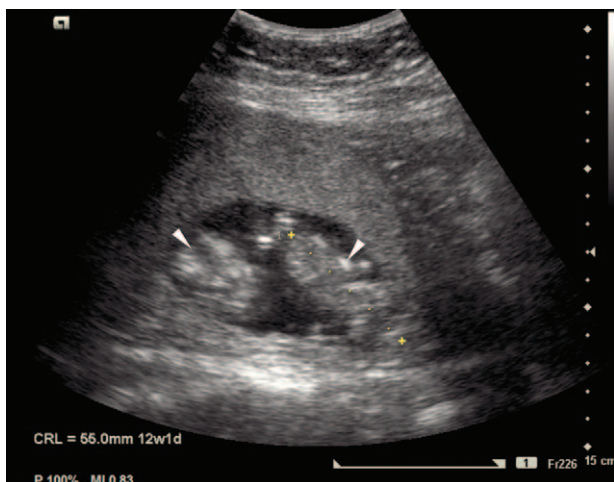


**Figure 2.** Transverse T2-weighted MRI with fat saturation showing empty uterine cavity (asterisk). MRI=magnetic resonance imaging.

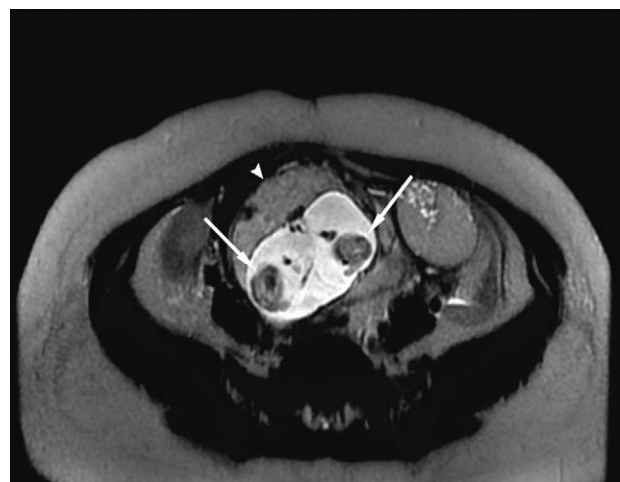
(~96%) in the Fallopian tubes.<sup>[6]</sup> It is the leading cause of maternal mortality in the first trimester, accounting for 4% to 10% of all pregnancy-related deaths.<sup>[7,8]</sup> This gynecological disorder has an incidence of 2% in the general population of pregnant women, but this rises to 16% in the ED.<sup>[4]</sup>

This case of ectopic pregnancy is particularly peculiar on 2 counts. First, it is a unilateral live spontaneous twin tubal ectopic pregnancy—similar cases are exceedingly rare, with very limited instances reported in medical literature worldwide thus far.<sup>[9]</sup> Second and more importantly, the patient presented on the cusp of her second trimester, much later than any of the prior reported cases. Such a presentation atypically late into the pregnancy represents a potential diagnostic pitfall in the ED, with EPs possibly missing such a diagnosis without MRI scans and placing the patient's life at risk.

A vast majority of patients with ectopic pregnancies present at a gestational age of 6 to 10 weeks, usually with symptoms of vaginal bleeding and/or abdominal pain.<sup>[5]</sup> They can be complicated with a rupture once the gestational sac grows



**Figure 1.** Transabdominal ultrasound demonstrating twin live pregnancy (arrowheads). The length of the foetal pole, that is, crown-rump length, measured 55.0mm corresponding to gestational age of 12 weeks 1 day.



**Figure 3.** Transverse single-shot fast spin-echo T2-weighted MRI revealing two sacs and fetuses (arrows) in right adnexa with placenta (arrowhead). MRI=magnetic resonance imaging.

beyond a diameter of 1.5 to 3.5 cm.<sup>[10,11]</sup> Pertaining to the subset of tubal ectopic pregnancies, unruptured and ruptured cases were found to present at gestational ages of  $6.9 \pm 1.9$  and  $7.2 \pm 2.2$  weeks, respectively.<sup>[12]</sup> This makes our patient's presentation of twin pregnancy at 12 weeks of gestation with amniotic sacs measuring 10 cm in diameter extremely rare and unusual.

The diagnosis of ectopic pregnancy in this patient was further concealed by her unremarkable medical history and physical examination findings. Based on the chief complaint of right iliac fossa pain in a pregnant lady, we included ectopic pregnancy and appendicitis as our 2 top differential diagnoses. There was an absence of risk factors for ectopic pregnancy save for advanced maternal age (age >35 years),<sup>[13]</sup> whereas physical and laboratory examination findings were equivocal for both diagnoses. Nevertheless, we opined that an ectopic pregnancy was improbable based on the patient's self-reported previous normal antenatal follow-ups and transabdominal ultrasonography revealing a viable live twin pregnancy.

In line with recommendations from the American College of Radiology for imaging in pregnant women, we then proceeded with MRI for further evaluation of our patient for appendicitis.<sup>[14–16]</sup> To our surprise, the gestational sac was localized at the right Fallopian tube with an empty uterine cavity, prompting the diagnosis of a live twin tubal ectopic pregnancy. On hindsight, the empty uterus visualized on MRI was probably obscured by the gestational sac and fetuses during ultrasonography. This demonstrates the potential discerning power of MRI in such ambiguous patients.

Early diagnosis of ectopic pregnancy is important due to the high mortality and morbidity risks associated with this condition. Tubal rupture and subsequent hypovolaemic shock occur in 32% of all such cases, with a 2.5% increase in risk for every 24 hours left untreated.<sup>[17]</sup> Urgency for early diagnosis and expedited treatment is even more pronounced in twin tubal pregnancies as seen in this case, which carry a 30% to 50% risk of hypovolaemic shock secondary to rupture.<sup>[18]</sup>

Our reported case thus illustrates a few key teaching points. First, EPs should not completely rule out the differential diagnosis of ectopic pregnancy solely based on prior normal antenatal examinations. Second, patients with tubal ectopic pregnancies can still present without rupture at gestational age of >10 weeks. Third, in the event of equivocal ultrasonography and laboratory findings, physicians should not hesitate to order MRI scans for further evaluation.

Spontaneous live unilateral tubal ectopic twin pregnancies are extremely rare, with prevalence estimated at 1 in 125,000 pregnancies.<sup>[19]</sup> Surgery is often the preferred mode of management in this subtype of ectopic pregnancies,<sup>[9]</sup> as was done for our patient.

#### 4. Conclusions

The diagnosis of ectopic pregnancy cannot be ruled out based on prior normal antenatal examinations and gestational age of >10 weeks. EPs should not hesitate to order MRI scans for further

evaluation if ultrasonography and laboratory findings are equivocal.

#### Author contributions

**Conceptualization:** Chen-June Seak, Zhong Ning Leonard Goh, Joanna Chen-Yeen Seak, Chen-Ken Seak.

**Resources:** Chen-June Seak.

**Supervision:** Chen-June Seak.

**Visualization:** Chen-June Seak.

**Writing – original draft:** Chen-June Seak, Alexis Ching Wong.

**Writing – review & editing:** Chen-June Seak, Zhong Ning Leonard Goh.

Chen-June Seak orcid: 0000-0003-0984-8385.

#### References

- [1] Cappell MS, Friedel D. Abdominal pain during pregnancy. *Gastroenterol Clin North Am* 2003;32:1–58.
- [2] El-Amin Ali M, Yahia Al-Shehri M, Zaki ZM, et al. Acute abdomen in pregnancy. *Int J Gynaecol Obstet* 1998;62:31–6.
- [3] Tamir IL, Bongard FS, Klein SR. Acute appendicitis in the pregnant patient. *Am J Surg* 1990;160:571–5. discussion 575–576.
- [4] Alkatout I, Honemeyer U, Strauss A, et al. Clinical diagnosis and treatment of ectopic pregnancy. *Obstet Gynecol Surv* 2013;68:571–81.
- [5] Murray H, Baakdah H, Bardell T, et al. Diagnosis and treatment of ectopic pregnancy. *CMAJ* 2005;173:905–12.
- [6] Bouyer J, Coste J, Fernandez H, et al. Sites of ectopic pregnancy: a 10 year population-based study of 1800 cases. *Hum Reprod* 2002;17:3224–30.
- [7] Creanga AA, Shapiro-Mendoza CK, Bish CL, et al. Trends in ectopic pregnancy mortality in the United States: 1980–2007. *Obstet Gynecol* 2011;117:837–43.
- [8] Berg CJ, Callaghan WM, Syverson C, et al. Pregnancy-related mortality in the United States, 1998 to 2005. *Obstet Gynecol* 2010;116:1302–9.
- [9] Kim C-I, Lee T-Y, Park S-T, et al. A rare case of spontaneous live unilateral twin tubal pregnancy with both fetuses presenting with heart activities and a literature review. *Obstet Gynecol Sci* 2018;61:274–7.
- [10] Gamzu R, Almog B, Levin Y, et al. The ultrasonographic appearance of tubal pregnancy in patients treated with methotrexate. *Hum Reprod* 2002;17:2585–7.
- [11] Balci O, Ozdemir S, Mahmoud AS, et al. The efficacy of multiple-dose methotrexate treatment for unruptured tubal ectopic pregnancy and conversion rate to surgery: a study on 294 cases. *Fertil Steril* 2010;93:2415–7.
- [12] Saxon D, Falcone T, Mascha EJ, et al. A study of ruptured tubal ectopic pregnancy. *Obstet Gynecol* 1997;90:46–9.
- [13] Farquhar CM. Ectopic pregnancy. *Lancet* 2005;366:583–91.
- [14] Garcia EM, Camacho MA, et al. Expert Panel on Gastrointestinal Imaging ACR Appropriateness Criteria® right lower quadrant pain-suspected appendicitis. *J Am Coll Radiol* 2018;15(11S):S373–87.
- [15] Kastenberg ZJ, Hurley MP, Luan A, et al. Cost-effectiveness of preoperative imaging for appendicitis after indeterminate ultrasonography in the second or third trimester of pregnancy. *Obstet Gynecol* 2013;122:821–9.
- [16] Theilen LH, Mellnick VM, Longman RE, et al. Utility of magnetic resonance imaging for suspected appendicitis in pregnant women. *Am J Obstet Gynecol* 2015;212:345.
- [17] Bickell NA, Bodian C, Anderson RM, et al. Time and the risk of ruptured tubal pregnancy. *Obstet Gynecol* 2004;104:789–94.
- [18] De Los Ríos JF, Castañeda JD, Miryam A. Bilateral ectopic pregnancy. *J Minim Invasive Gynecol* 2007;14:419–27.
- [19] Parker J, Hewson AD, Calder-Mason T, et al. Transvaginal ultrasound diagnosis of a live twin tubal ectopic pregnancy. *Australas Radiol* 1999;43:95–7.