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#### Contents

#### Thrice Monthly Volume 9 Number 23 August 16, 2021

#### **OPINION REVIEW**

	REVIEW
	Pérez Lara FJ, Jimenez Martinez MB, Pozo Muñoz F, Fontalba Navas A, Garcia Cisneros R, Garcia Larrosa MJ, Garcia Delgado I, Callejon Gil MDM
6582	COVID-19 pandemic, as experienced in the surgical service of a district hospital in Spain

6591 Beta-carotene and its protective effect on gastric cancer

Chen QH, Wu BK, Pan D, Sang LX, Chang B

Liver transplantation during global COVID-19 pandemic 6608

> Alfishawy M, Nso N, Nassar M, Ariyaratnam J, Bhuiyan S, Siddiqui RS, Li M, Chung H, Al Balakosy A, Alqassieh A, Fülöp T, Rizzo V, Daoud A, Soliman KM

6624 Nonalcoholic fatty pancreas disease: An emerging clinical challenge

Zhang CL, Wang JJ, Li JN, Yang Y

#### **MINIREVIEWS**

6639 Novel mechanism of hepatobiliary system damage and immunoglobulin G4 elevation caused by Clonorchis sinensis infection

Zhang XH, Huang D, Li YL, Chang B

6654 Intestinal microbiota participates in nonalcoholic fatty liver disease progression by affecting intestinal homeostasis

Zhang Y, Li JX, Zhang Y, Wang YL

- 6663 Theory and reality of antivirals against SARS-CoV-2 Zhao B, Yang TF, Zheng R
- 6674 Acute acalculous cholecystitis due to infectious causes Markaki I, Konsoula A, Markaki L, Spernovasilis N, Papadakis M

#### **ORIGINAL ARTICLE**

#### **Case Control Study**

Innate immunity - the hallmark of Helicobacter pylori infection in pediatric chronic gastritis 6686 Meliţ LE, Mărginean CO, Săsăran MO, Mocan S, Ghiga DV, Bogliş A, Duicu C

#### **Retrospective Study**

Effects on newborns of applying bupivacaine combined with different doses of fentanyl for cesarean 6698 section

Wang Y, Liu WX, Zhou XH, Yang M, Liu X, Zhang Y, Hai KR, Ye QS



<b>Contents</b> World Journal of Clinical Cas					
conter	Thrice Monthly Volume 9 Number 23 August 16, 202				
6705	Awake fiberoptic intubation and use of bronchial blockers in ankylosing spondylitis patients				
	Yang SZ, Huang SS, Yi WB, Lv WW, Li L, Qi F				
6717	Efficacy of different antibiotics in treatment of children with respiratory mycoplasma infection				
	Zhang MY, Zhao Y, Liu JF, Liu GP, Zhang RY, Wang LM				
6725	Expression of caspase-3 and hypoxia inducible factor $1\alpha$ in hepatocellular carcinoma complicated be hemorrhage and necrosis				
	Liang H, Wu JG, Wang F, Chen BX, Zou ST, Wang C, Luo SW				
6734	Increased morbidity and mortality of hepatocellular carcinoma patients in lower cost of living areas				
	Sempokuya T, Patel KP, Azawi M, Ma J, Wong LL				
	SYSTEMATIC REVIEWS				
6747	Safety of pancreatic surgery with special reference to antithrombotic therapy: A systematic review of the literature				
	Fujikawa T, Naito S				
6759	What paradigm shifts occurred in the management of acute diverticulitis during the COVID-19 pandemi A scoping review				
	Gallo G, Ortenzi M, Grossi U, Di Tanna GL, Pata F, Guerrieri M, Sammarco G, Di Saverio S				
	CASE REPORT				
6768	Pylephlebitis – a rare complication of a fish bone migration mimicking metastatic pancreatic cancer: case report				
	Bezerra S, França NJ, Mineiro F, Capela G, Duarte C, Mendes AR				
6775	Solitary seminal vesicle metastasis from ileal adenocarcinoma presenting with hematospermia: A cas				
	Cheng XB, Lu ZQ, Lam W, Yiu MK, Li JS				
6781	Hepatic abscess caused by esophageal foreign body misdiagnosed as cystadenocarcinoma by magnet resonance imaging: A case report				
	Pan W, Lin LJ, Meng ZW, Cai XR, Chen YL				
6789	2+0 CYP21A2 deletion carrier – a limitation of the genetic testing and counseling: A case report				
	Xi N, Song X, Wang XY, Qin SF, He GN, Sun LL, Chen XM				
6798	Psoriasis treatment using minimally manipulated umbilical cord-derived mesenchymal stem cells: A carreport				
	Ahn H, Lee SY, Jung WJ, Pi J, Lee KH				
6804	Double intussusception in a teenage child with Peutz-Jeghers syndrome: A case report				
0001					



World Journal of Clinical Cases						
Conter	nts Thrice Monthly Volume 9 Number 23 August 16, 2021					
6810	Nedaplatin-induced syndrome of inappropriate secretion of antidiuretic hormone: A case report and review of the literature					
	Tian L, He LY, Zhang HZ					
6816	Nasal metastases from neuroblastoma-a rare entity: Two case reports					
	Zhang Y, Guan WB, Wang RF, Yu WW, Jiang RQ, Liu Y, Wang LF, Wang J					
6824	Nocardiosis with diffuse involvement of the pleura: A case report					
	Wang P, Yi ML, Zhang CZ					
6832	Prenatal diagnosis of triphalangeal thumb-polysyndactyly syndrome by ultrasonography combined with genetic testing: A case report					
	Zhang SJ, Lin HB, Jiang QX, He SZ, Lyu GR					
6839	Blue LED as a new treatment to vaginal stenosis due pelvic radiotherapy: Two case reports					
	Barros D, Alvares C, Alencar T, Baqueiro P, Marianno A, Alves R, Lenzi J, Rezende LF, Lordelo P					
6846	Diverse microbiota in palatal radicular groove analyzed by Illumina sequencing: Four case reports					
	Tan XL, Chen X, Fu YJ, Ye L, Zhang L, Huang DM					
6858	Autism with dysphasia accompanied by mental retardation caused by <i>FOXP1</i> exon deletion: A case report					
0000	Lin SZ, Zhou XY, Wang WQ, Jiang K					
6867	FGFR2-TSC22D1, a novel FGFR2 fusion gene identified in a patient with colorectal cancer: A case report					
	Kao XM, Zhu X, Zhang JL, Chen SQ, Fan CG					
6872	Trismus originating from rare fungal myositis in pterygoid muscles: A case report					
	Bi L, Wei D, Wang B, He JF, Zhu HY, Wang HM					
6879	Retroperitoneal laparoscopic partial nephrectomy for unilateral synchronous multifocal renal carcinoma with different pathological types: A case report					
	Xiao YM, Yang SK, Wang Y, Mao D, Duan FL, Zhou SK					
6886	Diffuse large B cell lymphoma originating from the maxillary sinus with skin metastases: A case report and review of literature					
	Usuda D, Izumida T, Terada N, Sangen R, Higashikawa T, Sekiguchi S, Tanaka R, Suzuki M, Hotchi Y, Shimozawa S, Tokunaga S, Osugi I, Katou R, Ito S, Asako S, Takagi Y, Mishima K, Kondo A, Mizuno K, Takami H, Komatsu T, Oba J, Nomura T, Sugita M, Kasamaki Y					
6900	Manifestation of acute peritonitis and pneumonedema in scrub typhus without eschar: A case report					
	Zhou XL, Ye QL, Chen JQ, Li W, Dong HJ					
6907	Uterine tumor resembling an ovarian sex cord tumor: A case report and review of literature					
	Zhou FF, He YT, Li Y, Zhang M, Chen FH					
6916	Dopamine agonist responsive burning mouth syndrome: Report of eight cases					
	Du QC, Ge YY, Xiao WL, Wang WF					



Conton	World Journal of Clinical Cases
Conten	Thrice Monthly Volume 9 Number 23 August 16, 2021
6922	Complete withdrawal of glucocorticoids after dupilumab therapy in allergic bronchopulmonary aspergillosis: A case report
	Nishimura T, Okano T, Naito M, Tsuji C, Iwanaka S, Sakakura Y, Yasuma T, Fujimoto H, D'Alessandro-Gabazza CN, Oomoto Y, Kobayashi T, Gabazza EC, Ibata H
6929	Sirolimus treatment for neonate with blue rubber bleb nevus syndrome: A case report
	Yang SS, Yang M, Yue XJ, Tou JF
6935	Combined thoracoscopic and laparoscopic approach to remove a large retroperitoneal compound paraganglioma: A case report
	Liu C, Wen J, Li HZ, Ji ZG
6943	Menetrier's disease and differential diagnosis: A case report
	Wang HH, Zhao CC, Wang XL, Cheng ZN, Xie ZY
6950	Post-salpingectomy interstitial heterotopic pregnancy after <i>in vitro</i> fertilization and embryo transfer: A case report
	Wang Q, Pan XL, Qi XR
6956	Ulnar nerve injury associated with displaced distal radius fracture: Two case reports
	Yang JJ, Qu W, Wu YX, Jiang HJ



#### Contents

Thrice Monthly Volume 9 Number 23 August 16, 2021

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CASE REPORT

## Post-salpingectomy interstitial heterotopic pregnancy after in vitro fertilization and embryo transfer: A case report

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procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

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#### Abstract

#### BACKGROUND

Heterotopic pregnancy (HP) refers to the coexistence of ectopic pregnancy and intrauterine pregnancy. Salpingectomy is proposed as a pretreatment before in vitro fertilization and embryo transfer (IVF-ET) to reduce the risk of HP. HP after IVF-ET occurs in women who had already underwent bilateral salpingectomy, even though it is extremely rare.

#### CASE SUMMARY

A case of a 29-year-old woman with recurrent interstitial HP after IVF-ET following salpingectomy is presented. The main symptom was a sudden and worsening pelvic pain. Physical examinations revealed signs of peritoneal bleeding and irritation with stable vital signs. Transvaginal ultrasound showed a live intrauterine pregnancy and another live embryo with cardiac activity in the left cornu extending beyond the lateral edge of the uterus. Her hemoglobin concentration was 8.0 g/dL, and serum human chorionic gonadotropin value was 171116.9 mIU/mL. With the diagnosis of ruptured HP with internal bleeding, an emergency laparoscopic resection of left cornu was performed. The interstitial pregnancy was removed with caution to protect the intrauterine pregnancy. After the surgical treatment, the intrauterine pregnancy continued with no complications. A healthy baby was delivered by caesarean section at 39 wk. Outcomes of another three cases are further summarized.

#### CONCLUSION

Post-salpingectomy HP is a rare but challenging condition. Surgical treatment is preferred in the case with a viable intrauterine pregnancy.

Key Words: Heterotopic pregnancy; In vitro fertilization and embryo transfer; Salpingectomy; Surgery; Case report

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**Core Tip:** Our paper presented a rare case of a 29-year-old woman with recurrent interstitial heterotopic pregnancy (HP) after in vitro fertilization-embryo transfer following bilateral salpingectomy. We also summarized the outcomes of another three similar cases successfully treated in our department. Furthermore, we reviewed characteristics and outcomes of other reported post-salpingectomy HP cases. Interstitial HP after bilateral salpingectomy is extremely rare and has a high risk of rupture and hemorrhage.

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#### INTRODUCTION

Heterotopic pregnancy (HP) refers to the coexistence of ectopic pregnancy (EP) and intrauterine pregnancy and has a very low incidence ,occurring 1 in 30000 spontaneous pregnancies<sup>[1]</sup>. Due to the widespread use of assisted reproductive techniques, HP after in vitro fertilization and embryo transfer (IVF-ET) has been frequently reported<sup>[2]</sup>. Salpingectomy has been proposed as a pretreatment before IVF-ET to reduce the risk of HP. Theoretically, the whole visible part of the fallopian tube is removed by salpingectomy, which could prevent EP after IVF-ET. However, HP after IVF-ET even occurs in women who had already underwent bilateral salpingectomy.

Post-salpingectomy HP is an extremely rare but challenging condition, since the ectopic pregnancy must be removed whilst preserving the normal intrauterine pregnancy. Since the interstitial portion is rich in blood supply, it makes the postsalpingectomy interstitial HP an extremely dangerous complication with risks of sudden rupture and severe hemorrhage. Thus, for patients after tubal sterilization, the possibility of HP should not be neglected even with the presence of live intrauterine pregnancies. Previously reported post-salpingectomy HP cases have been reviewed[1-9]. Four of these cases were diagnosed as interstitial HPs (with three ruptured cases), only one of which reported a live birth of the intrauterine pregnancy after treatment. The other three cases experienced abortions.

Here, we report a case of recurrent and ruptured interstitial HP after IVF-ET following bilateral salpingectomy. We further summarize the outcomes and delivery modes of another three similar cases who were treated successfully in our department in the past 5 years.

#### CASE PRESENTATION

#### Chief complaints

A 29-year-old woman presented to our department complaining of worsening pain in the lower left abdominal quadrant. Her last menstrual cycle was 7 wk and 1 d previous.

#### History of present illness

The pain had started a day before her admission and had been worsened in the last 4 h. An IVF cycle was performed with two frozen embryos transferred 5 wk ago. There were no additional symptoms.

#### History of past illness

The patient had an obstetric history of gravida 4, para 0-0-3-0. Two years after her first spontaneous abortion, she twice underwent diagnostic hysteroscopy combined with laparoscopy for secondary infertility. Then, she had a history of HP with failing intrauterine pregnancy after ovulation induction treatment. Laparoscopic bilateral salpingectomy and dilation and curettage were performed. After sterilization, she



experienced two unsuccessful IVF cycles.

#### Personal and family history

There is no personal and family history.

#### Physical examination

The patient's temperature was 36.5 °C, heart rate was 95 bpm, respiratory rate was 20 breaths per min, blood pressure was 102/67 mmHg, and oxygen saturation was 98%. The general appearance was acute distress. The gynecologic examination revealed bulky uterus and tenderness in the left adnexal area, which showed signs of peritoneal bleeding and irritation but with stable vital signs.

#### Laboratory examinations

Blood analysis revealed an erythropenia  $(2.58 \times 10^{12}/L)$  and a hemoglobin concentration of 8.0 g/dL with normal leukocyte count and platelet count. Prothrombin and partial thromboplastin times were normal. Serum human chorionic gonadotropin value was 171116.9 mIU/mL. The blood biochemistries and urine analysis were normal.

#### Imaging examinations

Transvaginal ultrasound showed a live intrauterine pregnancy and another live embryo with cardiac activity in the left cornu extending beyond the lateral edge of the uterus. In addition, free fluid was noted in the cul-de-sac as well as in the abdomen.

#### FINAL DIAGNOSIS

The final diagnosis of the presented case is ruptured HP with internal bleeding after IVF-ET.

#### TREATMENT

An emergency laparoscopy was performed after discussion with the patient. After suction of blood clots and hemoperitoneum (about 700 mL), a ruptured interstitial pregnancy within the left salpingectomy site (lateral to the insertion of the left round ligament and not connected with uterine cavity) was confirmed (Figure 1). We performed a left cornual resection to remove the interstitial pregnancy, with caution to protect the intrauterine pregnancy.

#### OUTCOME AND FOLLOW-UP

The pathological report confirmed the presence of trophoblastic tissue in interstitial and intramural portion. The patient's post-operative course was uneventful. The synchronous intrauterine pregnancy continued with no complications and with appropriate growth of the fetus. A healthy baby was delivered by caesarean section at 39 wk, weighing 3110 g.

We further retrospectively studied cases of post-IVF interstitial HP following bilateral salpingectomy in our department over the past 5 years. The characteristics and outcomes of these 4 cases are listed in Table 1. All of them were treated by laparoscopic excision of the intramural and cornual segment and had very good outcomes for the intrauterine pregnancies.

#### DISCUSSION

As described above, salpingectomy or tube ligation is used to eliminate the risk of HP and improve outcomes of IVF-ET. The mechanism of HP following bilateral salpingectomy is unclear. The remaining interstitial portion of fallopian tube and the formation of a cornual sinus after laparoscopic salpingectomy might be the causative risk factors. Thus, it is important to perform an appropriate laparoscopic sterilization by minimizing tubal residue. Some studies further suggested cornual suture at the



Та	able 1 Characteristics of post-sterilization interstitial heterotopic pregnancy after in vitro fertilization and embryo transfer in our department											
No	Age, yr	Year	Gravida/Para	EP	History of sterilization	GA	Transferred embryos	Main symptom	Ultrasound manifestation	Ectopic site	Bleeding volume	Outcome of intrauterine pregnancy
1	29	2019	G4/P0030	1	Bilateral salpingectomy for HP in 2018	50 d	2 frozen embryos	Abdominal pain	IVE + ECM + PF	Left IP (ruptured)	700 mL	Full-term CS (selective)
2	32	2019	G4/P0030	2	Left salpingectomy for EP in 2013 and right salpingectomy for EP in 2015	66 d	2 fresh embryos	Abdominal pain	IVE + EVE	Left IP	300 mL	Premature CS at 34 wk (fetal distress)
3	36	2018	G4/P1021	1	Right salpingectomy and left tubal ligation for EP in 2015	38 d	2 frozen embryos	Abdominal pain	IVE + ECM + PF	Right IP (ruptured)	1000 mL	Full-term CS (breech position)
4	30	2016	G5/P0040	3	Left salpingectomy for EP in 2007 and right salpingectomy for EP in 2014	49 d	2 fresh embryos	None	IVE + EVE	Left IP	50 mL	Full-term CS (selective)

CS: Caesarean section; ECM: Extrauterine complex mass; EP: Ectopic pregnancy; EVE: Extrauterine viable embryo; GA: Gestational age; IP: Interstitial pregnancy; IVE: Intrauterine viable embryo; PF: Pelvic fluid.

time of salpingectomy to help reduce these risks[10].

HP can be treated by expectancy, medical or surgical treatment. If the intrauterine pregnancy is viable, surgical excision of the ectopic pregnancy is a better option. The intrauterine gestations in HP were more likely to end in spontaneous or induced abortion compared with the outcomes of normal intrauterine pregnancies. The rate of live birth after treatment was even lower. Thus, patients could be treated immediately and appropriately to increase the survival rate of the intrauterine pregnancy. In the case of a non-viable intrauterine pregnancy, conservative treatment should be considered if the patient is in stable condition without rupture. The treatment should be personalized.

Previously reported post-salpingectomy interstitial HP cases revealed a high rate of abortion. In the past 5 years, 4 cases of post-sterilization interstitial HP after IVF-ET were successfully treated by laparoscopic resection of the ectopic tissue and ipsilateral cornu in our department. Each of them had an ongoing healthy gestation culminating in a caesarean section with a live birth.

#### CONCLUSION

Post-salpingectomy interstitial HP should be considered as a separate diagnosis after IVF-ET. Timely diagnosis and immediate treatment are essential to achieve better clinical outcomes. In our experience, laparoscopy is the minimally invasive way to remove the ectopic tissue and preserve the viable intrauterine pregnancy with a very high success rate. Caesarean section is the preferred method of delivery considering the risk of metrorrhexis.

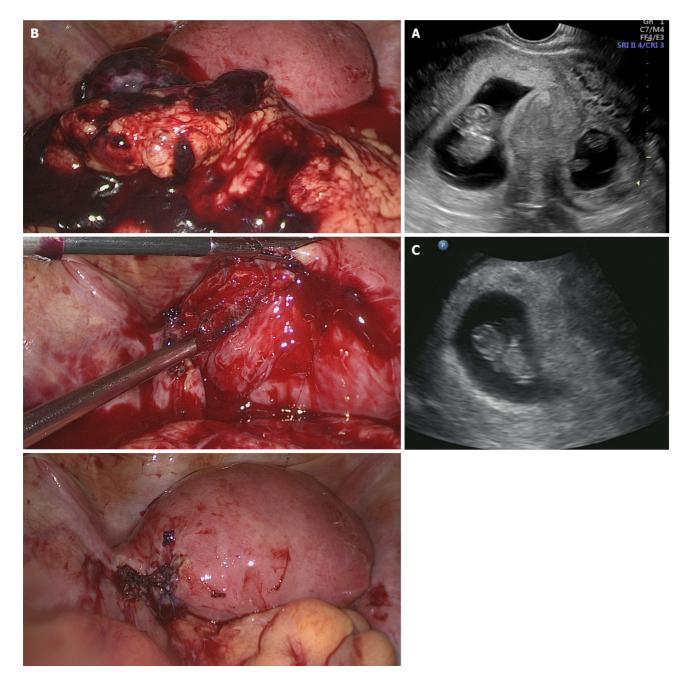


Figure 1 Transvaginal ultrasound images and laparoscopic excision of the intramural and cornual segment. A: Transvaginal ultrasound showed a live intrauterine pregnancy and another live embryo with cardiac activity in the left cornu and interstitial part; B: The patient was treated by laparoscopic excision of the intramural and cornual segment; C: Transvaginal ultrasound showed a healthy intrauterine fetus after surgery.

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