

Ectopic Pregnancy after Bilateral Tubal Ligation in a Woman with Asymptomatic Fitz-Hugh-Curtis Syndrome

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Abstract

This case report illustrates an incidental finding of asymptomatic Fitz-Hugh-Curtis syndrome in a 34-year old woman undergoing laparoscopy due to a spontaneous ruptured ectopic pregnancy after bilateral tubal ligation. To our knowledge, this is the first case to report an atypical presentation in a twofold aspect: the presence of perihepatic adhesions associated with asymptomatic pelvic inflammatory disease (Fitz-Hugh-Curtis Syndrome) with a history of bilateral tubal ligation, and the spontaneous tubal ectopic pregnancy after a failed tubal ligation. This report highlights the clinical importance of conducting an attentive examination of the pelvis via surgical exploration of all the abdominal cavity, including perihepatic space, especially in women with suspected pelvic inflammatory disease and ectopic pregnancy.

Keywords: Ectopic pregnancy, Fitz-Hugh-Curtis Syndrome, Laparoscopy, Atypical, Perihepatic Adhesions.

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BACKGROUND

The first case of Fitz-Hugh-Curtis syndrome (FHCS) was reported by Curtis et al. [1] and is characterized by perihepatic adhesions associated with pelvic inflammatory diseases (PID). Chlamydia trachomatis and Neisseria gonorrhoea have been identified as the main causative pathogens of PID and FHCS [2], with a prevalence of 12–14% among women of reproductive age [3]. The pathophysiology is attributed to haemato-lymphatic and peritoneal spread of pelvic

infections to the liver and hyper-immune response to C. trachomatis infection [4]. The management of FHCS includes the use of doxycycline and azithromycin for Chlamydia-associated FHCS [5]. The incidence of FHCS was found to be around 14% in women with ectopic pregnancy compared with 3% in those undergoing laparoscopic sterilization ($p < .01$) [6].

An ectopic pregnancy is an extra-uterine pregnancy. The aetiology often involves an inflammatory process located at the lumen or tissues surrounding the fallopian tubes. The incidence of ectopic pregnancy is strongly associated with an increased incidence of PID. Based on a retrospective, population-based cohort of women aged 15 to 44 years in Denmark (1995 to 2012), a positive test for

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chlamydia was positively associated with ectopic pregnancy (prevalence: 31%; adjusted hazard ratio: 1.31, 95% CI: 1.25–1.38), in comparison to women with a negative test for chlamydia [7].

The Collaborative Review of Sterilization (CREST) was a prospective, multi-centre study in the United States of 10,685 women between 1978 and 1987, demonstrating that female surgical sterilization is highly effective, and post-sterilization pregnancy occurs in <1% of procedures. Ectopic pregnancies accounted for 32.9% of all pregnancies reported in CREST subjects, with an ectopic pregnancy rate of 7.3 per 1000 in women who had undergone a sterilization procedure [7]. Multiple risk factors contribute to the development of ectopic pregnancy, particularly a history of sexually transmitted diseases and/or pelvic inflammatory disease. Sexual transmitted diseases (STD) are a major global health concern. The World Health Organization (WHO) estimated that the incidence of *C. trachomatis*, the most common STD, is 30 per 1,000 women, with an estimation that 30% will develop PID and tubal damage in the Middle East [8]. A population-based case-control study in Norway (n=616 participants) found that previous *C. trachomatis* infection was associated with elevated ectopic pregnancy risk (odds ratio: 1.4; 95% CI: 1.0–2.0) [9]. This finding was also reported in a prospective case-control study in Saudi Arabia (n= 135) stating that *C. trachomatis*, *M. genitalium*, and HSV-1/2 were more prevalent in ectopic pregnancy patients [10].

In a recent cohort study involving 30,450 patients with PID and 91,350 controls, the authors illustrated that patients with PID were more likely to develop ectopic pregnancy (cumulative incidence rate of 0.05%), and patients with PID had 2.121 times ($p=0.003$)

more risk of developing ectopic pregnancy than the controls [8]. Surprisingly, a recent report found that PID and ectopic pregnancy are risk factors for ovarian borderline tumours and low-grade serous carcinoma (LGSC) (HR: 1.95, 95% CI: 1.22–3.10; HR: 2.44, 95%: 1.20–4.96, respectively [9].

CASE PRESENTATION

A 34-year-old sexually active female was admitted to the emergency unit with intermittent abdominal pain for the last week, one episode of vaginal spotting, dizziness, fatigue, and missed period. Her medical history included bilateral tubal ligation a few years before. On initial examination, her vital signs were stable, apart from tachycardia. On palpation, her lower abdomen was mildly tender with mild rebound.

INVESTIGATIONS

Laboratory investigations showed increase in BHCG (>2000 IU/L) and high WBC and CRP count; the findings of ultrasound were suggestive of ectopic pregnancy with a mass seen towards the right adnexa, measuring approximately 7.8 x 3.9 cm, with free fluid and clots seen at the pouch of Douglas extending to the Morrison pouch (Figure 1a). The patient was hemodynamically stable but had low haemoglobin levels. The patient underwent emergency laparoscopy which revealed: hyperdense clots and intra-abdominal blood (around 1000 cc) with a ruptured right ampullary ectopic pregnancy that was 6–7cm in size (Figure 1b). Right salpingectomy with ligature was conducted, and haemostasis was achieved, with cleaning via pelvic irrigation. Omental and bowel adhesions were noted and dissected for specimen collection. During exploration of the perihepatic space, violin string-like adhesions

were noticed and Fitz-Hugh-Curtis syndrome was diagnosed (Figure 1c). The patient was treated postoperatively with IV cefuroxime and metronidazole for two days, then switched to doxycycline and Azithromycin for ten days, leading to satisfactory recovery. Histopathology microscopic description of the specimen showed fallopian tube luminal haemorrhage, hydropic chorionic villi and decidua, features consistent with an ectopic pregnancy. Some villi show trophoblastic proliferation and acute inflammatory cells.

DISCUSSION

To our knowledge, this is the first case of ruptured ectopic pregnancy after failed tubal ligation and with FHKS. Ectopic pregnancy should be suspected in any woman of reproductive age with abdominal pains, even after tubal ligation, despite fallopian tube ligation being a common, highly effective, and widely accepted permanent method of female contraception. However, should pregnancy occur following sterilization, the chances of ectopic are high. Several studies suggest that the incidence of ligation failure is 0.13–1.3%, of which 30–60% will be ectopic pregnancies [11].

Multiple risk factors contribute to the development of ectopic pregnancy, particularly a history of sexually transmitted diseases and/or pelvic inflammatory disease. STDs are a major health problem. The pooled prevalence of current genital infection in the Middle East is around 3.0% (95% CI: 2.3–3.8) in general populations 13.2% (95% CI: 7.2–20.7) in female sex workers, 11.3% (95% CI: 9.0–13.7) in infertility clinic attendees caused by PID, and 12.4% (95% CI: 9.4–15.7) in symptomatic women. Although the Middle East is burdened by *C. trachomatis* infection, the public health

response remains rudimentary and below WHO Global Health Sector Strategy standards on sexually transmitted syndromes [12].

Early diagnosis and treatment are essential in preventing the complications of ectopic pregnancy due to the high morbidity and mortality rate. It is a significant cause of first trimester maternal mortality (0.03 per 100,000), especially in women aged 25–34. As we did not include lysis in the consent form, we did not consider perihepatic adhesions, and hence we think that the inclusion of lysis of perihepatic adhesions in the preoperative consent form may be useful.

CONCLUSIONS

- 1- Tubal ectopic pregnancy can present with mild abdominal pain with subtle clinical findings, even after a bilateral tubal ligation.
- 2- There is an increased incidence of ectopic pregnancy in PID and Fitz-Hugh-Curtis syndrome.
- 3- Perihepatic space should be explored in all pelvic surgery, especially in those with PID and ectopic pregnancy.
- 4- The inclusion of lysis of perihepatic adhesions in the preoperative consent form in any woman suspected of ectopic pregnancy may be useful.

Contributors:

SM, HA & MA were involved in this patient's care. SM & MM contributed to the drafting of the manuscript. HA & MM contributed to the editing of the manuscript.

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FIGURE/VIDEO CAPTIONS

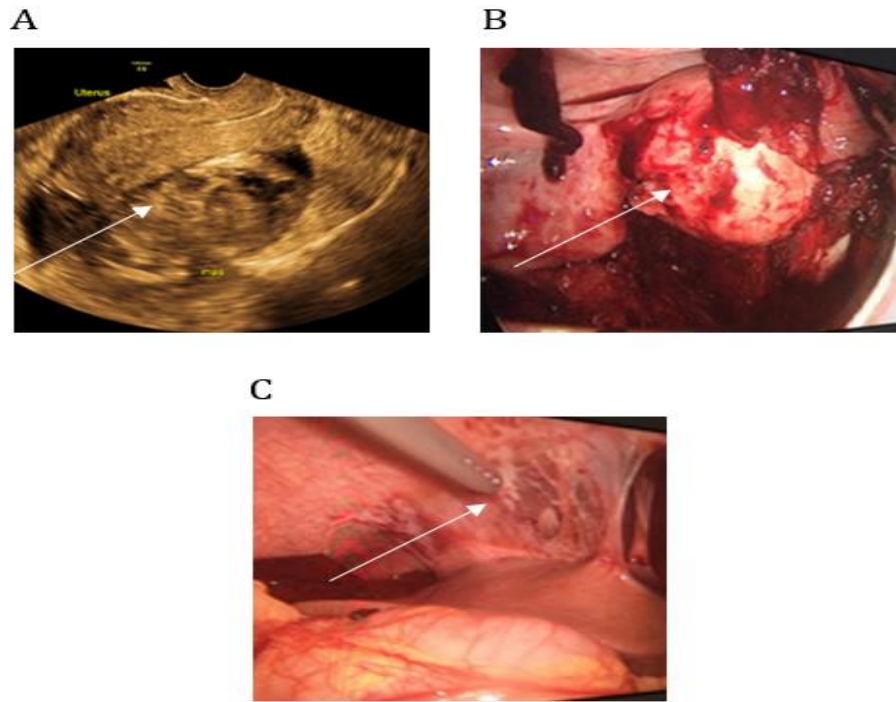


Figure 1: A- U/S scan showing right ectopic pregnancy adjacent to uterus (White arrow). B- Right ruptured ampullary ectopic pregnancy during laparoscopy (White arrow). C: Perihepatic adhesions and blood collection (White arrow).

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الحمل خارج الرحم بعد ربط أبقاق فالوب عند امرأة مصابة بمتلازمة فيتز-هيوغ-كورتيس بدون أعراض سريرية

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الملخص

توضح هذا التقرير عرضاً لمتلازمة فيتز-هيوغ-كيرتس غير المصحوبة بأعراض عند امرأة تبلغ من العمر (34) عامًا خضعت لتنظير البطن بسبب تمزق عفوي لحمل خارج الرحم بعد ربط الأنبوب الثنائي، وعلى حد علمنا فإن هذه الحالة الأولى التي أبلغت عن عرض غير نمطي في جانب مزدوج؛ وهو وجود التصاقات حول الكبد المرتبطة بمرض التهاب الحوض دون أعراض (متلازمة فيتز-هيوغ-كيرتس) مع ربط الأنبوب الثنائي المصاحب لحمل خارج الرحم، بعد فشل ربط البوقين، ويسلط هذا التقرير الضوء على الأهمية السريرية لإجراء فحص يقظ للحوض؛ من خلال الاستكشاف الجراحي لجميع تجاويف البطن بما في ذلك: الحيز حول الكبد، خاصة عند النساء اللواتي يشتبه عندهن أمراض التهاب الحوض المزمن، والحمل خارج الرحم.

الكلمات الدالة: الحمل خارج الرحم، متلازمة فيتز-هيوغ-كورتيس، أمراض التهاب الحوض المزمن، التنظير البطني.