

## CASE REPORT

# Fitz-Hugh-Curtis syndrome: An incidental diagnostic finding in an infertility workup

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## Abstract

A case of Fitz-Hugh-Curtis syndrome in a 32-year-old para 1<sup>+</sup> is reported. She presented with inability to conceive of 4 years duration. Her husband's semen analysis was within normal range. She had a hysterosalpingogram that showed bilateral tubal blockage and clinical assessment showed right sided abdominal tenderness, cervical excitation tenderness, and adnexa tenderness. The endocervical swab test for *Chlamydia trachomatis* was a positive. Laparoscopy and dye test showed adhesion bands on the under surface of the liver (the violin string appearance). She recovered well postoperatively. The couple received ofloxacin and metronidazole for 2 weeks. Literatures on Fitz-Hugh-Curtis syndrome presentation, pathogenesis, and management were reviewed.

**Key words:** Fitz-Hugh-Curtis syndrome, infertility, laparoscopy, pelvic inflammatory disease

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## Introduction

Fitz-Hugh-Curtis syndrome is a rare complication of pelvic inflammatory disease (PID) named after two physicians, Thomas Fitz Hugh Jr and Arthur Hale Curtis who first reported this condition in 1934 and 1930, respectively.<sup>[1,2]</sup> It was postulated that the syndrome was caused by *Neisseria gonorrhoeae* and *Chlamydia trachomatis* infections which cause thinning of cervical mucus and upward transmission of bacteria from the vagina to the uterus, fallopian tubes, paracolic gutter, and the liver capsule causing infection and inflammation.<sup>[3,4]</sup> Some evidence has demonstrated that the etiology of perihepatitis was due to a propulsion of peritoneal fluid from the pelvis to the diaphragm with preferential absorption on the right side.<sup>[5]</sup>

The exact pathophysiology/pathogenesis of this syndrome is still poorly understood, but there are several propositions ranging from direct spread, hematogenous spread, lymphatic spread, and exaggerated immune response. Currently, most experts believe that *C. trachomatis* is the culprit more than *N. gonorrhoeae*. There is a higher serum titer of anti-*Chlamydia* immunoglobulin G antibodies in patients with Fitz-Hugh-Curtis syndrome and salpingitis than in patients with salpingitis alone.<sup>[6-8]</sup> Perihepatitis and Fitz-Hugh-Curtis syndrome seem to represent a hyperimmune response to *C. trachomatis*.<sup>[6-8]</sup>

The syndrome is characterized by right upper quadrant pain, fever, abdominal pain, and vaginal discharge. Following an episode of PID, laparoscopy may reveal "violin string" adhesion of parietal peritoneum to the liver.

The management of Fitz-Hugh-Curtis syndrome is similar to that of PID.<sup>[9,10]</sup> In addition, the lysis of the adhesion bands

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may be done laparoscopically for symptomatic relief.<sup>[6,11]</sup> The long-term complications of Fitz-Hugh-Curtis syndrome are rare but are related to PID. They include abdominal pain, small bowel obstruction, and infertility.<sup>[11]</sup>

We, therefore, report a case of incidental findings of violin string appearance in a woman with infertility due to tubal factor on the background of PID following which Fitz-Hugh-Curtis syndrome was diagnosed. It is a rare finding which is associated with infertility and the organism commonly implicated is *C. trachomatis*.

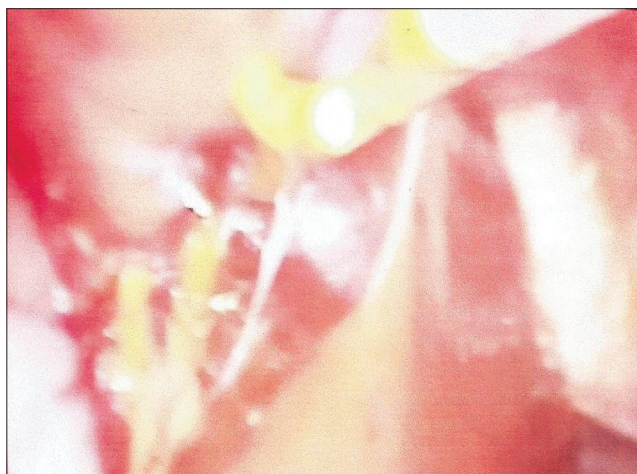
### Case Report

Mrs. C.I. a 32-year-old para 1<sup>+</sup> housewife who presented at our gynecological clinic with a complaint of inability to conceive of 4 years duration after marriage to a new husband despite regular unprotected sexual intercourse. Prior to the second marriage, she had a confinement in 2007 for the first husband, but they separated due to erectile dysfunction. She had a termination of unwanted pregnancy at 10 weeks gestation in 2009 for her male consort with no gross complication. There was a positive history of sexually transmitted infection and multiple sexual partners. She was living with her new husband and related well with him since marriage in 2010. There was a positive history of deep dyspareunia, dysmenorrhea, and low abdominal pain. There was no vaginal discharge or dysuria. There was no history of blurring of vision, persistent headache, heat or cold intolerance, galactorrhea, or undue loss of weight.

Husband had been assessed and his semen analysis was within normal range. She did a hysterosalpingogram which showed bilateral tubal blockage.

On examination, she was afebrile, anicteric and was not pale. Her height was 1.62 m and weight was 64 kg giving a body mass index of 24.4 kg/m<sup>2</sup>. The breasts were well-developed, and there was no palpable mass or expressible breast milk discharge. Her blood pressure was 130/70 mmHg and her pulse rate was 90 beats/min, regular and of good volume. Heart sounds I and II were heard with no murmurs. Her lung fields were clinically clear. Her abdomen was flat and moved with respiration. The umbilicus was inverted. There was abdominal tenderness on the right hypochondrium, epigastrium, and right Iliac fossa. The liver and spleen were not palpably enlarged. The kidneys were not ballotable. There were no masses felt. Bowel sound was present and normal. Vaginal examination showed female pattern of hair distribution. There was no vaginal discharge. Cervical excitation tenderness was positive and bimanual examination revealed normal-sized uterus that was not freely mobile.

A diagnosis of secondary infertility due to tubal factor was made and our findings were explained to her.



**Figure 1:** Violin string appearance on the under surface of the liver during laparoscopy in an infertility work up

She was booked for laparoscopy and dye test as well as hysteroscopy. The following investigations were done with results as shown in the following section.

- Packed cell volume: 38%
- Retroviral screening: Nonreactive
- Urinalysis: No abnormality detected
- Culture of peritoneal fluid aspirate: No growth of micro-organisms
- Acid fast bacilli test of peritoneal aspirate for *Mycobacterium tuberculosis*: Negative
- Gram stain of the endocervical swab specimen did not yield a Gram-negative diplococci
- Endocervical swab test for *C. trachomatis*: Positive
- Hysterosalpingogram showed bilateral tubal blockage.

Laparoscopy and dye test findings were adhesion bands on the under surface of the liver (violin string appearance) [Figure 1], bilateral hydrosalpinges with minimal spillage of dye from the adhesion bands under the surface of the right ovary; multiple adhesion bands on the right that held the right fallopian tube to the ovarian structure; dense adhesion bands on the left adnexa with no spillage. There was no gross abnormality on the other abdominal organs. Adhesiolysis was done on the right exposing the right tube. There was fluid in the pouch of Douglas measuring about 50 ml.

The findings were explained to them and the couple received ofloxacin 400 mg twice daily and metronidazole 400 mg trice daily for 2 weeks.

### Discussion

Fitz-Hugh-Curtis syndrome is a rare complication of PID. It occurs in 12.0–13.8% of pelvic inflammation cases.<sup>[12,13]</sup> The case reported had features of perihepatitis and chronic PID as there were tenderness on the right side of the abdomen and adhesions on the adnexae.<sup>[14]</sup> There was also the violin string appearance which is a typical evidence of the clinical features

of Fitz-Hugh-Curtis syndrome during laparoscopy. A definitive diagnosis can be made as was done in this case based on the detection of violin string-like adhesion or by identification of the causative organisms in hepatic capsular lesion specimen, which requires laparoscopy or laparotomy.<sup>[15-17]</sup> Perihepatitis capsular enhancement with the violin string appearance could be shown in other condition like peritonitis, liver abscess, acute cholecystitis, and carcinomatosis. These could be differentiated from Fitz-Hugh-Curtis syndrome using computer tomographic imaging. This was not done for our patient as it was not available in our setting.

*N. gonorrhoeae* and *C. trachomatis* were the common causative agents of Fitz-Hugh-Curtis syndrome<sup>[15]</sup> but currently most experts now report *C. trachomatis* as the more common causative pathogen.<sup>[15,17]</sup> Our findings confirmed *C. trachomatis* as the causative micro-organism for Fitz-Hugh-Curtis syndrome.

The first line of the management is to institute antibiotics sensitive to *C. trachomatis*.<sup>[15]</sup> Intravenous (IV) or oral regimen could be used according to guideline for treating PID as was done in this patient.<sup>[9,10]</sup> Our patient received oral antibiotics for *C. trachomatis*. In 1998, the centers for disease control and prevention published recommendations for the effective treatment of PID.<sup>[18]</sup> The first recommendation was ofloxacin 400 mg plus metronidazole 500 mg orally twice a day for 14 days as was done in the index case. Others included cefoxitin 2 g with probenecid 1 g orally concurrently, ceftriaxone 250 mg intramuscular (IM) (or other third generation cephalosporin such as ceftizoxime or cefotaxime) plus doxycycline 100 mg orally twice daily for 14 days.<sup>[18]</sup> In acute case parenteral treatment are the preferred recommendation and these included cefotetan 2 g IV 12 h or cefoxitin 2 g IV 6 h plus doxycycline 100 mg IV or orally 12 h, clindamycin 900 mg IV 8 h plus gentamicin IV or IM loading dose of 2 mg/kg body weight followed by a maintenance dose of 1.5 mg/kg 8 h (assuming normal renal function), ofloxacin 400 mg IV 12 h plus metronidazole 500 mg IV 8 h, ampicillin/sulbactam 3 g IV 6 h plus doxycycline 100 mg IV or orally 12 h or ciprofloxacin 200 mg IV 12 h plus doxycycline 100 mg IV or orally 12 h metronidazole 200 mg IV 8 h.<sup>[18]</sup>

All of these parenteral regimens should be continued until patients improvement is noted, and then doxycycline 100 mg orally twice a day should be continued for a total of 14 days of antibiotic treatment.<sup>[18]</sup>

In conclusion, Fitz-Hugh-Curtis syndrome should always be considered in the woman with infertility and the undersurface of the liver should always be visualized during

laparoscopy for tubal patency in all cases of infertility especially with background PID. *C. trachomatis* was seen as the causative micro-organism for Fitz-Hugh-Curtis syndrome. The consequences are that the organism implicated could damage the tubes progressively until it becomes irreparable and the recommendation is that antibiotic for PID could remedy the conditions before tubal damage.

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### Conflicts of interest

There are no conflicts of interest.

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