

Fitz Hue Curtis Syndrome; A Case Report and Literature Review

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Abstract

Background: Fitz Hugh Curtis Syndrome (FHCS) is a rare complication of pelvic inflammatory disease (PID) characterized by an inflammation of liver capsule without involvement of parenchyma with violin strings adhesion between the liver and adjacent structures. Mieg's syndrome is a triad of hydrothorax, ascites and ovarian fibroma. Cancer Antigen 125 is elevated in ovarian neoplasm. Ovarian cancer as well is the most common cause of omental cack. Up to our knowledge this is may be the first case report from the KSA.

Case Presentation: A Young unknown female who came to Saudi Arabia by illegal migrancy, presented to Emergency Department with progressive Shortness of Breath and gradual right upper quadrant pain for one month. She was undernourished thin black female, tachypneic, tachycardiac, desaturating, her abdomen was distended and tender. Laboratory results were leukocytosis, anemia, hypokalemia and hypomagnesaemia. Pleural and ascitic tapping was done, which showed a significant temporary clinical improvement. Chest X ray showed bilateral pleural effusion and an ultrasonography of abdomen concluded moderate ascites in subphrenic region and ruled out hepatobiliary problems. Abdomen Computed Tomography concluded omental cack, lymphadenopathy and fluid collection. The endoscopy revealed no active source of bleeding, with thickening of descending colon. Pelvic Ultrasonography showed free fluid and ovarian cystic mass with septation. Tumor markers were all normal except for Cancer Antigen 125 (CA125) was elevated. Mieg's syndrome was the clinically tangible diagnosis explained by the presence of hydrothorax, ascites, and ovarian mass. But did not explained the RUQ pain. Biphasic enhanced CT of abdomen and pelvis revealed early phase subcapsular enhancement, free fluid collection in the pelvis and abdomen, large cystic mass and tubule-ovarian abscess. FHCS diagnosis was established and laparotomy was done where the patient improved and was discharge on empirical antibiotic.

Conclusion: FHCS is a vague and multisystemic condition but once is diagnosed, treatment is well-known and improvement to fast. However, early pick up of the diagnosis is integral to put paid a permanent disability and undesirable fate. To our knowledge this was the first case to be reported from the Kingdom of Saudi Arabia.

Keywords: *Fitz-Hugh-Curtis syndrome (FHCS); Pelvic Inflammatory Disease (PID); RUQ Pain Ascites; Peritonitis; Pleural Effusion; Peri-hepatitis; Laparotomy*

Abbreviations

FHCS: Fitz Hugh Curtis Syndrome; PID: Pelvic Inflammatory Disease

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Introduction

Fitz Hugh Curtis Syndrome (FHCS), perihepatitis, is a rare complication of pelvic inflammatory disease (PID). It is characterized as an inflammation of the liver capsule that spared the parenchyma and associated with typical violin-string adhesions between liver and adjacent structure [1].

FHCS was first illustrated by Stajano in non-English publication in 1920 [1]. Ten years later, Arther Curtis Hale described an adhesion between the anterior surface of the liver and abdominal wall found during laparotomies of cases presented with right upper quadrant pain [1]. In 1934, Thomas Fitz Hugh jr, described similar cases with unusual localized peritonitis, involving the anterior surface and edge of liver and adjacent peritoneal surface of diaphragm [1-3].

FHCS is usually affecting a woman of childbearing age who is sexually active [1,4,5]. A notable number of cases were reported amongst male gender, though [6-10]. FCHS is very rare complicating only from 4 to 27 percent of PID [11-14]. The latter is commonly due to chlamydial or gonococcal infection [7,15-17]. *Chlamydia trachomatis* is the most common pathogen for FCHS worldwide [15,16]. *Neisseria gonorrhoeae* was also not uncommon [4,7,16]. Furthermore, other pathogens, such as *Mycobacteria tuberculosis* [16,18] and nontuberculous *Mycobacteria* [19] were reported in some cases.

FHCS is presenting a diagnostic challenge as it has a vagarious clinical vignette that mimicking many other more common differential diagnoses and involving many body systems [14,15]. Based on current literature the main clinical presentation which was frequently reported is the right upper quadrant pain, precluding dozens of differential diagnoses, such as hepatobiliary diseases, hepatitis, hepatic abscess, peritonitis, pyelonephritis, appendicitis, enterocolitis, and so forth [13,15,17,20]. These conditions were the reasoning behind the incidental discovery of the violin-string adhesions consistent with FHCS by invasive investigation modalities such as laparoscopy and/or laparotomy [20-23]. In spite of being complex and tough to pick, FHCS is completely curative once diagnosed [3,8,10,17-21,23]. In addition, the clinical outcome depends on at which stage of disease the diagnosis was established. To clarify, some permanent complications such as infertility may ensue at late stage of the disease [18,22]. Therefore, we came to the conclusion that in spite of the masked vague clinical presentation, FHCS is curable condition and the early establishment of diagnosis put limit to undesirable consequences.

As time passes, the clinical alertness about the condition and the importance of its diagnosis increased giving rise to a highly sensitive and specific non-invasive modality, namely biphasic enhanced-computed tomography superseded the need invasive procedures [5,14,17,20].

Here we are presenting a critical case of ovarian mass associated with FHCS which was admitted to Intensive Care Unit requiring oxygen supplement, vasopressor and inotropic support, blood transfusion and other resuscitation measures. It was really critical and had been deteriorating, going from pallor to post, and was on the verge of succumbing to the illness until the establishment of the diagnosis of FCHS and Mieg's syndrome.

Case Presentation

Young unknown female presented to Emergency Department with gradual Right Upper Quadrant pain for 5 weeks and progressive shortness of breath for 1 month. Shortness of breath is progressive interfering with minimal activity but not associated with Paroxysmal nocturnal dyspnea, orthopnea, or cough. The pain is localized stabbing abdominal pain in increasing with inspiration and movement radiating to the right shoulder and inside of right arm. On physical examination she was found to be desaturated (O_2 sat = 88 - 89 on room air), tachypneic (RR = 25 - 28 bpm), tachycardiac (HR = 115 - 120 bpm) but normotensive (88/48 mmHg) and afebrile (temperature was 38.8°C). Chest auscultation revealed bilateral equal air entry diminished peripherally but no rhonchi or wheezing, Cardiovascular system concluded normal heart sound no friction rub or muffled heart sound no raised JVP. Abdominal was distended dull to percussion dullness more appreciated centrally. The rest of exam was unremarkable.

Chest X-ray revealed massive right pleural effusion as well as abdominal ultrasound showed mild- moderate ascites. Pleural tapping was done where the right lung re-gain its elasticity and compliance swiftly return to completely normal status. Ascitic tapping and in-and-out drainage was done.

Complete blood count showed leukocytosis, normocytic normochromic anemia with very low hemoglobin level (6.2 g/dl) prompting blood transfusion, chemistry and serum electrolytes showed hypokalemia and hypomagnesaemia. Patient was admitted to ICU isolation bed as a case of sepsis/septic shock, norepinephrine 0.05 mcg/kg/minute, empirical antibiotic started, oxygen supplement with non-re-breather mask 15 liters was set, one-unit packed RBC was transfused after controlling her fever, electrolytes were replaced and screening for sepsis, HIV, COVID-19 and AFB were sent and came out to be negative for all.

Computed tomography (CT) of abdomen showed thickness of the descending colon, omental cack. Upper and lower endoscopic examination did not figure any source of bleeding.

At this stage we saw it imperative to dig deep in history and repeat the clinical assessment accordingly. We came out with the following clinical features. She is a young lady medically free had a gradual right upper quadrant pain and episodes of shortness of breath. She added that she is currently suffering a neck and right arm pain started two days ago. Clinical examination again revealed nothing apart from the aforementioned. Here we came to the conclusion that we should include the genitourinary system problems as a valid source of bleeding, and an explanation of her case.

Pregnancy test and Tumor markers were all requested came out to be negative except for CA125 was elevated. Pelvic ultrasound was done to rule out ectopic pregnancy and ovarian problems revealing pelvic free fluid a large cystic pelvic lesion with septate. This prompted the need of CT with contrast of both abdomen and pelvic which concluded the presence of tubule-ovarian abscess, bilateral salpingitis, pelvic free fluids and early enhancement of the hepatic capsule.

The pleural effusion was more in the right lung and it was resolved completely after drainage. No sooner than the lung had regained its normal status and the respiratory distress disappeared the formation of another pleural fluid started again. In addition, the fluid drained from the percutaneous intercostal tail was serious transudate by definition of light. This is actually hydrothorax not the pleural effusion. The ascites was gelatinous yellowish thick fluid with Serum ascitic protein ratio more than. The anemia was not explained by all radiological investigations this alert us to figure out another possible source of bleeding. Pelvic ultrasound showed large cystic mass with septation. Mieg's syndrome triad was therefore clinically evident namely hydrothorax, ascites, and ovarian fibroma.

Patients was diagnosed as FHCS and she underwent Ex. laparotomy for excision and biopsy for the pelvic mass adhesions identification and subsequent adhenolysis. There were and adhesions between the liver and diaphragm and anterior surface of liver to peritoneal cavity (or abdominal wall). These adhesions could've explained the right massive pleural effusions and ascites rather than Meigs syndrome.

Appropriate coverage of the possible microorganisms with dual antibiotic regimen was started patient improved clinically and discharged from the hospital after one week.

Results and Discussion

With respect of the current literature Fitz Hugh Curtis Syndrome (FHCS) is a diagnostic challenge and need to be included as differential diagnosis of right upper abdominal pain especially in women of child-bearing age who are sexually active [5,14,15,17,18,20,24,25]. Although, some authors reported that it should be raised as a differential diagnosis of RUQ pain regardless of the gender, although female is the typical presentation [4-7,9,10]. FHCS is attributed to PID and this is the reasoning behind assuming it to be a female-prevailing disease. Rather, it was reported amongst male gender who presented with RUQ pain and CT findings consistent with FHCS as well as

Chlamydia trachomatis isolated from urine. Thus, ascribing the condition to an infectious pathogen is more precise and wide-ranging than describing it as a complication of PID. Or on second thought, it is worth mentioning that FHCS is a complication of infection from the pathogens that are usually isolated from cases of both PID and STD. These are *Chlamydia trachomatis* and *Neisseria gonorrhoea* in the most part of time other microorganisms has been isolated and reported with every-changing literature.

The presence of tubule-ovarian abscess, salpingitis, free fluid in pelvic and abdominal cavities, ascites, pleural effusion, increased CA125 omental cack, pelvic mass, and mesenteric lymphadenopathy were the new findings in this case and were not previously known to be consistent with FHCS. These findings could be rearranged in organized sequence of occurrence as PID, (i.e., evident as Salpingitis tubule-ovarian abscess free fluid collection), complicated by FHCS, as reported by Biphasic CT scan and as it was tangible in laparotomy findings violin string adhesions of liver capsule to adjacent structures namely diaphragm and peritoneum. The latter caused the pleural effusion which mainly in Right lung, and ascites.

What is open to questioning is the presence of pelvic mass, elevated CA120, and mesenteric lymphadenopathy. Along with Ascites and Pleural effusions constructed the triad of meig's syndrome. Meig's syndrome is composed of ovarian fibroma, ascites and hydrothorax. Honestly, the nature of the pleural effusion in this case goes well with hydrothorax as it is transudate, improved tremendously after in/out Percutaneous tail insertion and drainage. Rather, it was unilateral right pleural effusion. It is reported with FHCS cases right pleural effusion and attribute to the adhesion between the liver capsule to the adjacent right side of diaphragm [26]. An elevated Ca125 was previously elaborated among PID cases [27]. Other studies suggested that elevated CA120 is indicative and prognostic parameter of tuberculosis peritonitis [28,29].

Patient reported a concomitant features of neck pain and right arm which were reported amongst cases of FHCS previously [30].

As a matter of fact, the diagnosis of FHCS was first suggested by radiologist who noticed a continuation of pelvic fluid collection through-out the abdominal cavity approaching the perihepatic region. Then Biphasic Enhanced Computed Tomography was done and showed Hyperenhancement of liver capsule.

Conclusion

According to our knowledge it was the first FHCS case reported from our country. It was intricated clinical syndrome that led to ICU admission, exploited advance imaging and investigations, consumption of procedures and treatment which all could be inappropriate. Once the definite diagnosis was established, the appropriate medical and surgical intervention were done the case improved incredibly and the patient's fertility was preserved. Thus, Fitz hue Curtis Syndrome is curable once diagnosed and it could be halted from progressing towards undesirable fate such as infertility.

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Conflict of Interest

Authors disclose no conflicts of interest.

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