Rupture of Dermoid Cyst- Unusual Cause of Fitz-Hugh-Curtis Syndrome

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Introduction
Fitz-Hugh-Curtis (FHC) syndrome is characterized by acute right upper quadrant pain and perihepatitis, typically following pelvic inflammatory disease (PID). 1, 2 It was first described in the 1930s by Thomas Fitz-Hugh and Arthur Curtis, who reported “violin string” adhesions between the liver and the abdominal wall in patients with gonococcal salpingitis. 3-5 Direct visualization of FHC syndrome with laparoscopy or laparotomy shows purulent and fibrinous exudates on the liver surface acutely and capsular thickening and adhesions in the chronic phase. 1, 2 WBC count, ESR and liver enzymes are elevated in some patients, although hepatic parenchymal involvement is minimal. 6, 7 FHC syndrome can be rarely caused by other etiologies, including systemic lupus erythematosi, IUD insertion, endometriosis, raltegravir and post-renal transplant with mycoplasma hominis infection. 8-16 Only one other case of post-operative perihepatitis has been published, describing a complication following a laparoscopic tubal ligation. 17

Case Presentation
A 41-year-old female underwent laparoscopic cystectomy for a right ovarian dermoid cyst that was complicated by intraoperative cyst rupture and spillage of a copious amount of sebaceous material and hair. Despite vigorous irrigation of the peritoneal cavity, a small amount of spilled cyst content remained at the end of the surgery. Laparoscopic examination of the upper abdomen revealed no adhesions and normal bowel. Pathology of the surgical specimen demonstrated a dermoid cyst containing skin, adnexal, and smooth muscle components, as well as an incidental 1 cm carcinoid tumour in the cyst wall. The patient denied diarrhea, headaches, sweats or any other symptoms associated with a carcinoid tumour.

The patient’s post-operative course was complicated by bilateral pulmonary embolism on day 7. A CT scan of the thorax demonstrated bibasilar atelectasis and a right pleural effusion in addition to pulmonary emboli. Laboratory investigations revealed an elevated WBC count at 19.9 x 10^6 (Normal 4 – 11 x 10^6). All other laboratory findings including liver function tests, bilirubin, and blood and urine cultures were normal. A follow-up CT scan performed two months later demonstrated a marked increase in the size of the right pleural effusion and a new complex perihepatic collection (Figure 1).

As part of the treatment for the carcinoid tumor, laparoscopic right salpingo-oophorectomy and appendectomy was performed three months following the initial cystectomy. Examination under laparoscopy confirmed the presence of Fitz-Hugh-Curtis adhesions around both lobes of the liver and adhesions within the pelvis. The patient tolerated the surgery well, and remained asymptomatic post-operatively. Final pathology showed removal of the residual teratoma, with negative peritoneal fluid cytology and cultures. Abdominal MRI performed four months following the initial surgery showed a persistent multiloculated and septated perihepatic collection centered in the right subphrenic region demonstrating enhancement of septae and restricted diffusion (Figure 2). In addition, diffusion-weighted imaging (DWI) demonstrated restricted diffusion along the caudate lobe in the porta hepatis, and peri-pancreatic and paraaortic regions (Figure 3). A CT scan of the abdomen one month later demonstrated persistent loculated fluid in the perihepatic, peripancreatic, and porta hepatis regions.

Figure 1. Axial (A) and sagittal (B) CT images demonstrate a right pleural effusion, high density perihepatic fluid and thickening around the liver.
reproductive age, MR has become a valuable modality in defining abdominal pathology in this population. However, the manifestation of perihepatitis on MR imaging remains inadequately characterized. One of the few MR studies on perihepatitis showed a thickened hepatic capsule of slightly increased signal intensity on T2-weighted imaging (T2-WI) and patchy enhancement of the liver parenchyma on portal venous phase imaging. In our case, T2-WI demonstrated hyperintense perihepatic collections but did not show inflammatory adhesions in the porta hepatis, and peri-pancreatic and para-aortic regions. However, these features were identified by DWI. Previous literature has shown improved characterization of focal liver lesions with the combination of conventional MRI and DWI as compared with conventional MR alone. DWI has also proven its use in identifying and characterizing non-oncological pathologies of the liver, in particular the assessment of liver fibrosis and cirrhosis.

We had confirmation of FHC adhesions on laparoscopy, and thus were able to differentiate from other causes of hepatic capsular enhancement. However, the appearance of hepatic capsular enhancement or perihepatic complex fluid on MRI should also alert radiologists to the differential diagnosis of perforated cholecystitis, perforated hepatic abscess, tuberculous peritonitis, pseudomyxoma peritonei, and peritoneal metastases from GI and ovarian tumours.

Characteristic CT and MR findings of perihepatic enhancement can direct towards a diagnosis of Fitz-Hugh-Curtis (FHC) syndrome without symptomatic presentation. Added value of DWI is in identifying inflammatory lesions otherwise inconspicuous on T2-weighted imaging. Finally, the case should bring to the attention of surgeons and radiologists that intraoperative cyst rupture can lead to the rare complication of FHC syndrome.

**Discussion**

Our patient presented with an atypical cause of FHC syndrome, secondary to intraoperative dermoid cyst rupture. The syndrome was found incidentally on CT scan and subsequently confirmed by intra-abdominal visualization by laparoscopy. It is possible that the symptoms of the disease, in the form of pleuritic right upper quadrant abdominal pain, were missed due to concurrent presentation of pulmonary embolism. The right pleural effusion was initially thought to be caused by acute pulmonary embolism; however, perihepatitis has been reported to occur with right pleural effusion. Our patient was likely diagnosed in the chronic phase and the symptoms regressed without requiring treatment.

This case emphasizes the importance of radiographic findings in diagnosing atypical causes of perihepatitis, independent of clinical symptomatology and/or isolation of a characteristic pathogen. Traditionally, the non-specific appearance of perihepatic fluid collection and less often fibrous bands on ultrasound have been used to corroborate clinical suspicion of FHC syndrome. In recent years, CT imaging showing hepatic capsular enhancement on arterial phase has been established to be a sensitive and efficient diagnostic modality.

Given the importance of minimizing radiation exposure in women of