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Case report

Fitz-Hugh-Curtis syndrome with disseminated intra-abdominal gonorrhoea

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ABSTRACT

Neisseria gonorrhoea continues to be implicated in a large proportion of sexually transmitted infections worldwide. Prompt recognition of infection is required to prevent further complications which include pelvic inflammatory disease and less commonly, perihepatitis which is known eponymously as Fitz-Hugh-Curtis syndrome. Third generation cephalosporins such as ceftriaxone remain effective in the treatment of gonococcal infection, however failure in initiation of appropriate antibiotic therapy in a timely manner can result in further disseminated disease. We describe an atypical case of Fitz-Hugh-Curtis syndrome presenting with multiple intra-abdominal gonococcal collections. Our case highlights the importance of a detailed sexual history in the evaluation of acute abdominal pain in at-risk patient demographics.

Introduction

Despite continuous advancements in public health and antimicrobial therapies, sexually transmitted infections (STI) continue to pose significant challenges worldwide. *Neisseria gonorrhoea*, a gram-negative diplococcus, has an estimated incidence of 87.9 million cases worldwide per year [1]. *N. gonorrhoea* infection classically presents with urogenital sequelae, including dysuria and urethral discharge, though many individuals remain asymptomatic [2]. Gonorrhoeal infection can also manifest as a localised septic arthritis and as an arthritis-dermatitis syndrome [3]. Other rare extragenital complications have been described, including perihepatitis, also known as Fitz-Hugh-Curtis syndrome (FHCS), gonococcal meningitis and infective endocarditis [4–6].

FHCS was first documented in 1930 and has subsequently gained increasing recognition as a rare complication of pelvic inflammatory disease (PID) in which *N. gonorrhoea* is commonly implicated [4,7,8]. The classic presentation of FHCS typically involves systemically-well females of childbearing age who present with acute atypical right upper quadrant abdominal pain which is often mistaken for other hepatobiliary or gastrointestinal pathology [4,7,9,10]. Radiological findings in FHCS are often quite subtle. In contrast, we present a severe and atypical case of FHCS with disseminated intra-abdominal gonococcal infection and associated systemic inflammatory response syndrome requiring initial inotropic and vasopressor support.

Case report

A 35-year-old female re-presented to a metropolitan emergency department with persistent epigastric and subdiaphragmatic pleuritic pain without nausea or vomiting. Medical history was unremarkable apart from previous dilatation and curettage in the context of endometriosis. The patient denied any previous abdominal surgeries and had no known family history of inflammatory bowel disease. She was not taking any regular medications. A binge type pattern of alcohol consumption and infrequent cocaine use was volunteered.

The patient had been discharged two days earlier from another tertiary hospital, following a 10-day long admission, where she had been admitted for acute lower abdominal pain with associated systemic inflammatory response syndrome. During her index admission, there were no symptoms suggestive of an acute urogenital or gastrointestinal infection. Blood tests had initially revealed a significant inflammatory picture: white cell count 26.2×10^9 L ($3.9–12.7 \times 10^9$ L), platelets 288×10^9/L ($150–450 \times 10^9$/L), neutrophils 24.2×10^9 L ($1.9–8.0 \times 10^9$ L) and C-reactive protein (CRP) 157 mg/L (0–5 mg/L). She required a brief intensive care unit admission for ionotropic support (intravenous noradrenaline) due to persistent hypotension. A viral infective screen,

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which included serological assay for human immunodeficiency virus and hepatitis A and B serology, was performed, and was negative. A general septic screen including a mid-stream urine sample and multiple blood cultures did not isolate a causative agent. Computed tomography (CT) imaging of the abdomen revealed thick oedematous small bowel loops with associated omental stranding (Fig. 1A) and a moderate amount of free fluid (Fig. 1B), however no evidence of ischaemia or visceral perforation was noted. A seven-day course of intravenous amoxicillin-clavulanic acid and a further three-day oral tail was completed prior to discharge. A presumptive diagnosis of cocaine-induced mesenteric ischaemia was made, and the patient was discharged on the basis of resolving abdominal pain requiring simple analgesia. Her white cell count and CRP on discharge were 19.7×10^9 L and 121 mg/L respectively. On repeat presentation, she was alert and haemodynamically stable but was clinically mildly hypovolaemic. The patient had visible discomfort on deep inspiration, though lung fields were clear on auscultation. Abdominal examination was notable for generalised tenderness; most prominently in the right iliac fossa and paraumbilical region, without signs of peritonism. Bowel sounds were present and active. Full blood examination demonstrated a haemoglobin of 100 g/L (115–160 g/L) with moderate rouleux formation, a white cell count of 22.2×10^9/L with a predominant neutrophilia and a platelet count of 1473×10^9/L with an elevated C-reactive protein 56 mg/L (<5 mg/L). Liver function testing and coagulation studies were within normal limits apart from an isolated hypoalbuminemia 25 g/L (33–46 g/L). Renal function was within normal range as were electrolytes. Lipase levels were elevated at 300 units/L (10–55 units/L). Quantitative beta-human chorionic gonadotrophin was negative.

Given the marked thrombocytosis and subdiaphragmatic pleuritic chest pain, a CT pulmonary angiogram was performed to excluded pulmonary embolism. This study was unremarkable. A contrast-enhanced CT abdomen and pelvis was also performed and revealed collections in the pouch of Douglas with thick capsule formation (Fig. 2A), as well as in the suprapubic region (Fig. 2 A), right middle abdomen (Fig. 2B), right paracolic gutter and left pelvic region. Increased perihepatic enhancement was also demonstrated (Fig. 3). The patient was monitored closely, without antimicrobial therapy, for several days, and both her neutrophil count and CRP gradually fell. Intra-abdominal drain tubes were inserted with diagnostic and therapeutic intent, but were removed shortly after insertion due to significant associated pain. Fluid aspirate was sent for microscopy, culture and sensitivity testing in addition to polymerase chain reaction for sexually transmissible infections. There was a marked leukocytosis with nil bacterial growth. However, polymerase chain reaction analysis of the drain fluid, as well as a subsequent first pass urine sample, returned positive for *N. gonorrhoea*. A diagnosis of FHCS with disseminated intra-abdominal gonococcal infection was made given the exclusion of other infective aetiologies, characteristic perihepatic enhancement on CT imaging and demonstration of *N. gonorrhoea* infection. A pelvic ultrasound was performed to evaluate for complications of PID which demonstrated normal tubo-ovarian anatomy. Testing for other common sexually transmitted infections were negative. A single dose of intravenous ceftriaxone was administered, and a ten-day course of oral doxycycline prescribed to be completed post discharge. Monthly follow-up was organised for the first three months, with repeat CT abdomen and pelvis imaging two-months from initial discharge demonstrating a marked reduction in the abdominal collections and no residual pelvic collections. The patient remained well. Testing of previous sexual contacts was recommended.

Discussion

Historically, diagnosis of FHCS was made by direct visualisation of characteristic “violin string” adhesions between the anterior liver surface and abdominal wall or diaphragm during diagnostic laparoscopy or laparotomy in the appropriate clinical context [7,11]. Due to radiological advances, a diagnosis of FHCS can now be made based on a high index of clinical suspicion together with evidence of perihepatic enhancement on computed tomography or magnetic resonance imaging following the exclusion of other relevant pathologies [4,11–13]. FHCS is typically considered as a complication or extension of PID due to its predominance in women of childbearing age, in which *N. gonorrhoea* and *Chlamydia trachomatis* are commonly implicated [10,13]. It has been proposed that ascending infection from the upper genital tract results in trans-fallopian spread into the peritoneal cavity and paracolic gutters prior to making contact with the liver capsule [7,11]. It is now appreciated that translocation of various microorganisms involved in FHCS can also occur via haematogenous or lymphatic spread, which is evidenced by known cases of FHCS in males [10,11,14,15]. Notwithstanding this, intra-abdominal collections with systemic inflammatory response syndrome is very rare in the context of FHCS. A constellation of atypical history, examination and investigation features led to assessment for *N. gonorrhoea* and *Chlamydia trachomatis* in our patient. Specifically, the predominant symptom of subdiaphragmatic pain, the improvement in inflammatory markers without initial anti-microbial re-treatment, and the finding of intra-abdominal collections on serial CT imaging without overt peritonism, or evidence of ancillary underlying bowel pathology. A retrospective review of the patient’s index CT revealed a small amount of non-encapsulated fluid around the dome of the liver, in the subhepatic space, in both paracolic gutters, and in the pouch of Douglas. In the right clinical context, this may have pointed to FHCS as a differential for atypical abdominal pain. On serial CT, 12-days later, the collections had increased markedly in volume, with formation of a thick capsule. The radiological and clinical findings were suggestive of FHCS, with perihepatitis and peritonitis and fluid collections with florid inflammation causing thick capsule formation. In terms of treatment, current CDC guidelines recommend a single dose 500 mg intramuscular ceftriaxone and further cover for concurrent Chlamydia

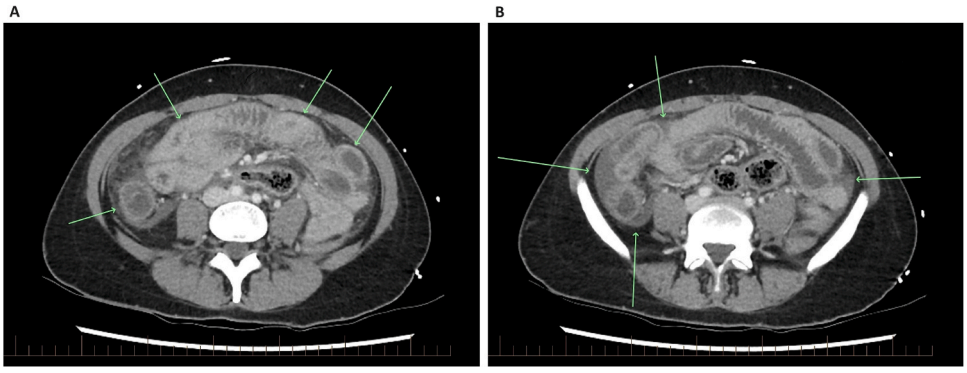

Fig. 1. Title – Contrast-enhanced CT abdomen and pelvis on initial presentation. Description – CT abdomen and pelvis demonstrating thickened oedematous small bowel loops (A) and moderate free fluid accumulation (B).

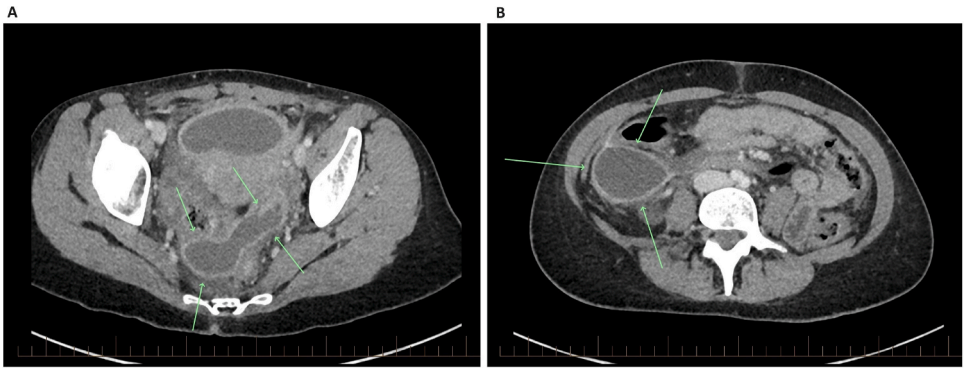

Fig. 2. Title – Contrast-enhanced CT abdomen and pelvis on re-presentation. Description – Extensive intra-abdominal collections with thick encapsulated collections in the pouch of Douglas and suprapublic region (A) and right paracolic gutter (B).

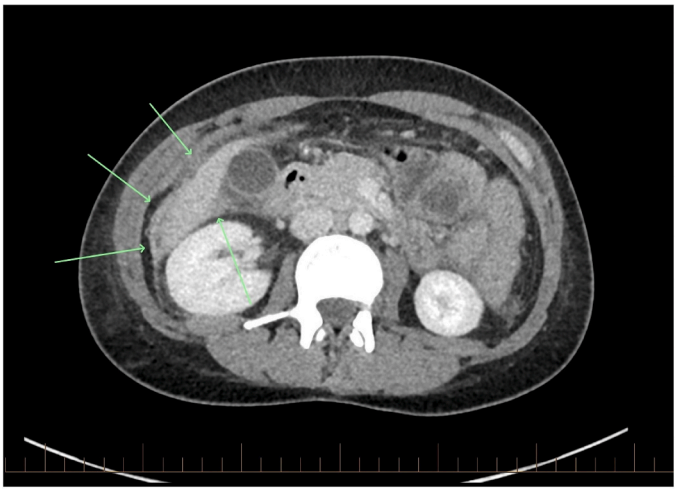

Fig. 3. Title – Contrast-enhanced CT abdomen and pelvis demonstrating Fitz-Hugh-Curtis syndrome. Description – Characteristic perihepatic enhancement on contrast-enhanced CT of the abdomen and pelvis suggestive of Fitz-Hugh-Curtis syndrome.

infection (if not excluded) with oral 100 mg doxycycline twice daily for seven days for uncomplicated urogenital gonococcal infection [16]. For disseminated gonococcal infection manifesting as septic arthritis or an arthritis-dermatitis syndrome, intravenous or intramuscular ceftriaxone 1 g daily is recommended prior to susceptibility testing for a total course of seven days, however no specific recommendations are made for intra-abdominal infection/collections [16]. Of note, antibiotic resistance amongst strains of *N. gonorrhoea* has been increasingly noted with demonstration of resistance to penicillin, ciprofloxacin, azithromycin and even third generation cephalosporins [16,17]. It is thus possible that the initial use of amoxicillin-clavulanic acid in our patient was insufficient to mitigate disease progression. In summary, we illustrate an atypical case of FHCS presenting with a systemic inflammatory response syndrome and disseminated intra-abdominal gonococcal infection. This case emphasises the importance of considering sexually transmitted infections as a potential cause of undifferentiated acute abdominal pain in at-risk populations. It is also imperative to obtain a detailed sexual history, including an evaluation of high-risk behaviours in patients with severe abdominal pain where an acute surgical cause has been excluded. The case also highlights the importance of multidisciplinary evaluation in patients with atypical abdominal pain, particularly given that findings from the index CT imaging may have raised suspicion of FHCS in the right clinical context. Suspicion of STI should prompt investigation with a first-pass urine analysis to allow for timely and targeted antimicrobial therapy. In doing so, complications including pelvic inflammatory disease and disseminated gonococcal infection could potentially be mitigated.

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Author statement

All authors have contributed to the manuscript in terms of concept, writing and editing. All authors were primarily responsible for the concept and design and were part of the treating team. RT, AWN and JHA were responsible for the draft of the manuscript. All authors were responsible for critical revision for important intellectual content and editing of the manuscript and have given final approval of the article for publication. All authors read and approved the final manuscript.

CRediT authorship contribution statement

Violette Cohen-Hallaleh: Formal analysis, Investigation. **Jeremy Druce:** Data curation, Formal analysis, Investigation. **Joshua Haron Abasszade:** Data curation, Formal analysis, Project administration, Writing – review & editing. **Andrew William Nguyen:** Data curation, Writing – original draft. **Michael Braude:** Conceptualization, Data curation, Formal analysis, Investigation, Project administration, Supervision, Writing – review & editing. **Ruyi Tan:** Project administration, Writing – original draft, Writing – review & editing.

Declaration of Competing Interest

The authors declare that there are no conflicts of interest regarding the publication of this article.

Data availability statement

The participant of this study did not give written consent for their data to be shared publicly, so due to the sensitive nature of the research, supporting data is not available.

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