INTRODUCTION

Pelvic inflammatory disease (PID) is a cause of infertility but is often not scrutinized in patients with abdominal pain. Fitz-Hugh-Curtis syndrome (FHCS) is perihepatitis associated with genital tract infections including Chlamydia trachomatis. It accounts for as much as one fourth of cases with PID. We report an unusual unique case of appendicitis that was complicated by Chlamydia trachomatis-induced FHCS and cervicitis. Differential diagnosis is difficult because the appendix is anatomically proximal to the right-side fallopian tube and because both etiologies can occur simultaneously.

CASE PRESENTATION

A 22-year-old Japanese woman came to our hospital after 2 days of vomiting and 3 h of pain in the lower abdomen that worsened with walking. Her medical history included bipolar disorder. She was a university student with a history of unprotected sexual intercourse with multiple partners. At the time of presentation, she had unprotected sexual intercourse with multiple partners. She had high Alvarado score and lack of cervical motion pain, despite cervical inflammation. Noncontrast CT showed enlarged appendix. Laparoscopic appendectomy revealed acute suppurative appendicitis and perihepatic adhesion. Cervical PCR assay was positive for C. trachomatis. She remained febrile but defervesced after azithromycin therapy. Clinicians should confirm whether females with abdominal pain are sexually active in view of screening for C. trachomatis.

KEYWORDS
appendicitis, Carnett's sign, Fitz-Hugh-Curtis syndrome, pelvic inflammatory disease
indicating a problem within the abdominal cavity. Pelvic examination revealed bloody vaginal discharge and an inflamed fornix uteri with no cervical motion pain and adnexal tenderness on bimanual examination. Polymerase chain reaction (PCR) tests of the cervical swab sample against *Neisseria gonorrhoeae* and *Chlamydia trachomatis* and a culture test for bacteria were sent to the laboratory.

Laboratory examination revealed an elevated C-reactive protein of 15.2 mg/dl, elevated white blood cell counts of 12,180/μl) and 86.9% neutrophils (normal range: 42%-77%) with an Alvarado score of 8. The levels of alanine transaminase and aspartate transaminase were 20 and 21 U/L, respectively. Urinary beta human chorionic gonadotropin (β-hCG) test was negative. Noncontrast abdominal computed tomography (CT) showed an enlarged appendix with surrounding fat stranding (Figure 1). With provisional clinical diagnosis of acute appendicitis, a general surgeon was consulted without performance contrast CT. The patient was admitted as an inpatient.

Laparoscopic appendectomy revealed inflammation of the entire wall of the appendix with a small amount of bloody ascites in the Douglas fossa, which was consistent with acute suppurative appendicitis. Adhesion was observed in the superior ileoceleal fold, and there was perihepatic adhesion. Intravenous ampicillin-sulbactam was administered, but her fever did not subside, even after 3 days of treatment. Figure 2 shows the patient’s course of therapy. Cervical PCR assay was found to be positive for *C. trachomatis* and negative for *N. gonorrhea*. The patient was therefore diagnosed as having *C. trachomatis*-induced FHCS. She took azithromycin and deferred the next day. Epigastric pain during testing for Carnett’s sign gradually subsided. Pelvic examination showed normal fornix uteri, and PCR for *C. trachomatis* was negative after 3 weeks. On our recommendation, her partner was tested for *C. trachomatis* by PCR assay, which was found to be negative. The patient has provided permission to publish the features of her case, and her identity has been protected.

### DISCUSSION

*Chlamydia trachomatis* has been reported as a cause of secondary appendicitis. In cases diagnosed with PID, appendicitis was found in 3.4%. We suspected appendicitis in this case from the patient’s high Alvarado score, family history of appendicitis, and lack of cervical motion pain. Her sexual history and malodorous vaginal discharge led us to consider PID, including pelvic examination and subsequent PCR screening. Cervical motion pain is a key physical finding suggestive of PID in evaluation of acute pelvic pain. In predicting PID, it has high sensitivity (82%) and moderate specificity (72%). Evaluation for PID as well as appendicitis is also recommended, especially in sexually active women, even those without cervical motion pain.

To the best of our knowledge, only three cases of FHCS accompanied by appendicitis have been reported. Joshi et al reported gonococcal FHCS with pathological evidence of appendicitis and peritonitis. Kazama et al reported culture-negative FHCS with radiological evidence of appendicitis. Hamdan et al reported FHCS in a man with ectopic appendicitis. Meanwhile, the currently reported case is of chlamydial FHCS in a woman with appendicitis. Joshi et al diagnosed FHCS clinically (fever, abdominal pain, elevated transaminase level and *N. gonorrhoeae* yielded from peritoneal pus), whereas Kazama et al made clinical and radiological diagnosis of FHCS (fever, abdominal pain, elevated transaminase level and CT finding of hepatic capsular enhancement in the arterial phase). Unlike in the case reported by Kazama et al, our patient lacked right upper abdominal tenderness. Besides acute fever and abdominal pain, the diagnostic findings of FHCS in our case were positive Carnett’s sign over the epigastric area and perihepatic
adhesion confirmed by laparoscopic surgery with positive PCR of the cervical swab sample for *C. trachomatis*. Carnett’s sign is a useful physical examination for distinguishing abdominal wall pain from visceral pain. Kijima et al. reported that it is also useful to detect adhesion-induced abdominal pain. Meanwhile, Carnett’s sign can also be positive in patients with appendicitis. The value of single physical finding should not be overestimated, however, and should be interpreted cautiously with the clinical course, especially in cases with intercurrent appendicitis. We prioritized management of appendicitis and made a surgical consultation after taking non-contrast CT. Our patient defervesced, and the epigastric pain was no longer shown in Carnett’s test after administration of azithromycin. Cervicitis was later resolved, and *C. trachomatis* was confirmed by PCR response.

It is unclear in our patient whether appendicitis occurred secondary to PID or concurrently with PID. Besides cervical swab PCR for *C. trachomatis*, PCR test in the appendix would have revealed the role of *C. trachomatis* in the pathogenesis of appendicitis in our patient. Cervical swab PCR was sent to a remote laboratory, and so the lag time for the results of PCR was a limitation of our institute. On site rapid multiplex PCR assay for pathogens known to cause PID might facilitate quicker investigation of both etiologies.

In conclusion, FHCS and PID are rarely concurrent with appendicitis but can be a differential diagnosis of appendicitis, especially if they experience any symptoms of PID.

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**CONFLICT OF INTEREST**

The authors have stated explicitly that there are no conflicts of interest in connection with this article.

**REFERENCES**

1. Paavonen J. Immunopathogenesis of pelvic inflammatory disease and infertility – what do we know and what shall we do? J Br Fer Soc. 1996;1(1):42–5.
2. Peter NG, Clark LR, Jaeger JR. Fitz-Hugh-Curtis syndrome: a diagnosis to consider in women with right upper quadrant pain. Clevel Clin J Med. 2004;71(3):233–9.
3. Mårth PA, Walner-Hanssen P. Periappendicitis and chlamydial salpingitis. Surg Gynecol Obstet. 1985;160(4):304–6.
4. Kahn JG, Walker CK, Washington AE, Landers DV, Sweet RL. Diagnosing pelvic inflammatory disease: a comprehensive analysis and considerations for developing a new model. JAMA. 1991;266(18):2594–604.
5. Joshi RM, Alkhalegy AA. Acute gonococcal Fitz-Hugh-Curtis syndrome: a case report. Int J STD AIDS. 2012;23(3):e39–40.
6. Kazama I, Nakajima T. A case of fitz-hugh-curtis syndrome complicated by appendicitis conservatively treated with antibiotics. Clin Med Insights Case Rep. 2013;6:35–40.
7. Hamdan M, Johane H, Benhamou G. The Fitz-Hugh-Curtis syndrome in a man revealed by ectopic appendicitis. Eur J Med. 1992;1(5):314–5.
8. Suleiman S, Johnston DE. The abdominal wall: an overlooked source of pain. Am Fam Physician. 2001;64(3):431–8.
9. Kijima T, Hyakudomi R, Hashimoto T, Kusaka A, Nakatani T, Ishibashi Y. Adhesion-induced chronic abdominal pain: a case report on the diagnostic value of Carnett’s test. J Med Case Rep. 2019;13(1):93.
10. Gray DW, Dixon JM, Seabrook G, Collin J. Is abdominal wall tenderness a useful sign in the diagnosis of non-specific abdominal pain? Ann R Coll Surg Engl. 1988;70(4):233–4.

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