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Case Report

An Uncommon Manifestation of Fitz-Hugh-Curtis Syndrome with Right-side Chest Pain

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Fitz-Hugh-Curtis syndrome is characterized by an inflammation of the perihepatic capsules associated with pelvic inflammatory disease. The typical symptom is severe right upper quadrant abdominal pain. We report a patient with Fitz-Hugh-Curtis syndrome who presented with an atypical chief complaint of right-side chest pain unaccompanied by symptoms specific to pelvic inflammatory disease. This case indicates that Fitz-Hugh-Curtis syndrome should be considered in the differential diagnosis of right-side chest pain in young women, because early diagnosis and treatment of the disease are essential to prevent chronic complications.

Key words: Fitz-Hugh-Curtis syndrome (FHCS), pleurisy, right-side chest pain

Fitz-Hugh-Curtis syndrome (FHCS) is an inflammation of the liver capsule occurring as a complication of pelvic inflammatory disease (PID), and occurs mainly as a result of accompanying *Chlamydia trachomatis* infection [1, 2]. Most patients are women of childbearing age who seek treatment for acute pain in the right upper abdomen, occasionally accompanied by lower abdominal pain or abnormal vaginal discharge [3, 4]. We hereby report a rare case of FHCS with a chief complaint of right-side chest pain without any pelvic inflammatory symptoms.

Case Presentation

A 20-year-old woman presented to our hospital with a chief complaint of right-side chest pain. She had no significant medical history or any relevant family history. Five days prior to visiting our hospital, she suddenly developed right lower chest pain and a fever with a body temperature of up to 37.4℃, for which she visited her family physician. Abdominal ultrasonography and plain computed tomography (CT) of the chest and abdomen revealed no abnormalities. The family physician prescribed loxoprofen and cefdinir. Two days prior to her visit to our hospital, she visited her family physician again as her chest pain persisted. Table 1 shows the results of laboratory studies performed on that day. Upon her visit to our hospital, she was first referred to a breast surgeon, whose physical examination of her breast revealed no significant findings. The patient was then referred to our department on the same day.

She had a spontaneous stabbing, knocking pain in her right lower chest, and the pain was most severe at the seventh intercostal space of the anterior axillary line (Fig. 1). The pain was exacerbated by deep breathing and twisting of the upper body. She denied having lower abdominal pain, vaginal discharge, or dysuria. Her menstrual period started 10 days prior

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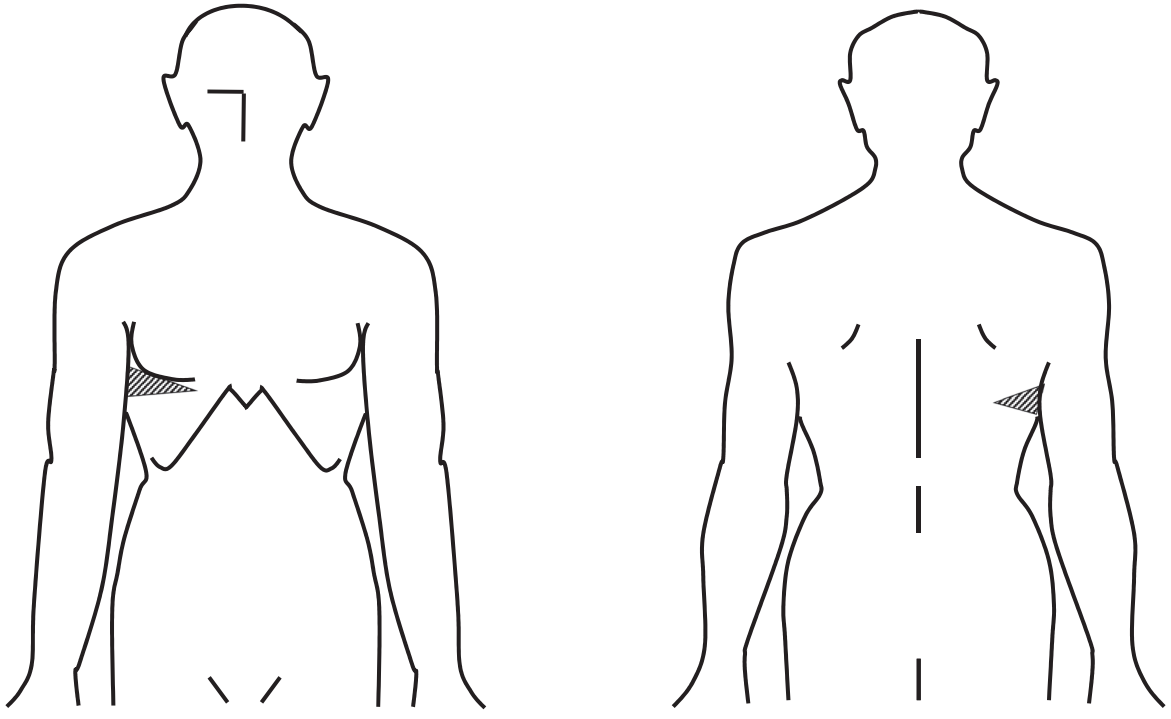

Fig. 1 The pain was localized in the seventh intercostal space of the anterior axillary line (shaded area).

Table 1 Laboratory data

Complete blood count		Blood chemistry serology	
Red blood cell	$421 \times 10^4/\mu l$	T-Bil	0.13 mg/dl
Hemoglobin	12.8 g/dl	AST	15 U/l
Hematocrit	38.0 %	ALT	12 U/l
White blood cell	9,300/μl	ALP	200 U/l
Neutrophil	69.1 %	γ-GTP	11 U/l
Lymphocyte	20.2 %	LDH	134 U/l
Monocyte	7.5 %	Na	140 mEq/l
Basophil	0.4 %	K	5.0 mEq/l
Eosinophil	2.8 %	CK	33 U/l
Platelets	$37.0 \times 10^4/\mu l$	CRP	3.83 mg/dl

to her visit to our hospital. Her sexual history included only one male sexual partner, but detailed information, such as frequency or the last date of sexual intercourse, was not obtained from the medical interview. In our physical examination, her blood pressure was 130/74 mmHg, heart rate 67 beats/min, and regular, and axillary body temperature 36.9℃. Her lungs were clear to auscultation bilaterally, and her heart sounds were regular, without any cardiac murmurs or rubs. Her abdomen was flat, without tenderness or Murphy's sign. Neurological examination revealed no abnormalities.

Laboratory findings are shown in Table 2. They revealed the following values: white blood cells, 8,220/μl with 74.6% neutrophils; C-reactive protein, 1.82 mg/dl; and erythrocyte sedimentation rate in 1 h, 74 mm. Considering her age and pleuritic pain, serum levels of antinuclear antibodies and complement activity were measured in order to rule out systemic lupus erythematosus and were found to be within their respective normal limits. An enhanced computed tomography (CT) demonstrated a slightly enhanced middle-to-right hepatic surface, suggesting inflammation of perihepatic capsules (Fig. 2). The amount of pleural effusion was within the normal limit. Although the patient did not complain of any abdominal symptoms, FHCS was suspected because of the CT findings. Serological studies yielded positive results for both serum *C. trachomatis* immunoglobulin (Ig) A and IgG. The patient was referred to a gynecologist on the same day of her visit. An internal examination found no tenderness in her adnexa and Douglas' pouch, and transvaginal ultrasonography revealed a slightly increased amount of ascites. A polymerase chain reaction (PCR) study of the gene amplification of a cervi-

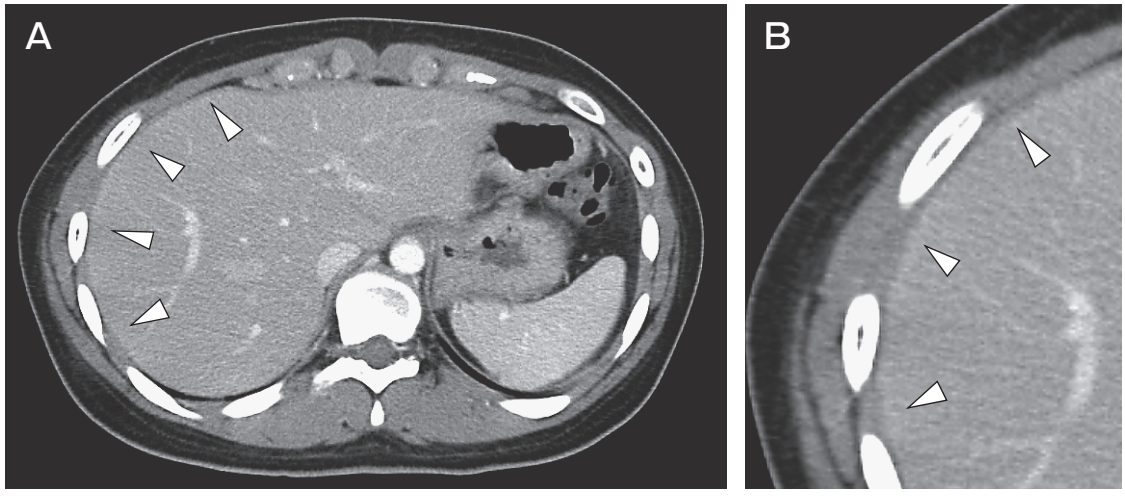

Fig. 2 Contrast-enhanced CT image of the abdomen demonstrating a slightly enhanced hepatic surface (arrows).

Table 2 Laboratory data

Complete blood count		Blood chemistry serology			
Red blood cell	$416 \times 10^4/\mu l$	T-Bil	0.22 mg/dl	RPR (syphilis)	(−)
Hemoglobin	12.2 g/dl	AST	13 U/l	TPLA (syphilis)	(−)
Hematocrit	36.6 %	ALT	1 U/l	HIV Ab	(−)
White blood cell	8,220/μl	ALP	204 U/l	HBs Ab	(−)
Neutrophil	74.6 %	γ-GTP	9 U/l	HBs Ag	(−)
Lymphocyte	20.1 %	LDH	151 U/l	HCV Ab	(−)
Monocyte	3.6 %	BUN	12.3 mg/dl	Anti-*Chlamydia trachomatis* IgG	(+)5.41
Basophil	0.4 %	Cr	0.50 mg/dl	Anti-*C. trachomatis* IgA	(+) > 10
Eosinophil	1.4 %	Na	139 mEq/l	PCR (*C. trachomatis*)	(+)
Platelets	$42.1 \times 10^4/\mu l$	K	4.4 mEq/l	PCR (*Neisseria gonorrhoeae*)	(−)
		Cl	104 mEq/l		
		Ca	9.3 mg/dl	Urinalysis	
		CK	38 U/l	pH	7.0
		T-protein	7.9 g/dl	Protein	(−)
		Albumin	3.7 g/dl	Occult blood	(−)
		CRP	1.82 mg/dl	Glucose	(−)
		ESR	74 mm	White blood cell	3/HPF

RPR, rapid plasma regain; TPLA, *Treponema pallidum* latex agglutination; PCR, polymerase chain reaction

cal mucus sample was positive for *C. trachomatis*. Other serological tests were performed to rule out sexually transmitted diseases, including human immunodeficiency virus infection, hepatitis B virus infection, hepatitis C virus infection, syphilis, and *Neisseria gonorrhoeae* infection. All of the test results were negative. Although there was no clear evidence of ascending infection, the increased amount of ascites suggested the possibility of PID. Also, the positive results for *C. trachomatis* strongly indicated a diagnosis of FHCS. Treatment was initiated with a single oral dose of 2-g/day azithromycin, which was also given to her sexual partner. Her right-side chest pain gradually decreased, disappearing completely 4 days after treatment initiation. This clinical course also supported the possibility that her symptoms were caused by FHCS due to *C. trachomatis* infection.

Discussion

FHCS is characterized by inflammation of the perihepatic capsules associated with pelvic inflamma-

tion. The most common causative agent is *C. trachomatis*, which causes perihepatitis associated with "violin string" adhesion between the hepatic surface, diaphragm, and abdominal wall. Perihepatitis is directly visualized only by diagnostic laparoscopy or laparotomy; however, both of these are invasive, and thus neither is frequently performed [2]. Therefore, in a clinical setting, the diagnosis can be adequately established by the findings of enhanced CT scanning, isolation of characteristic pathogens such as *C. trachomatis* or *Neisseria gonorrhoeae*, and exclusion of other causes [1]. In our case, pleuritis, pneumonia, and pneumothorax were considered as differential diagnoses because the patient was a young woman and her chief complaint was a pleuritic chest pain [5]. However, all of these diagnoses were unlikely, judging from the results of laboratory studies and the CT findings. In addition, a slightly enhanced hepatic surface with enhanced CT, and the positive results of *C. trachomatis* in serological studies and PCR, strongly supported the diagnosis of FHCS in our case.

Because the typical symptoms of FHCS include sudden onset of sharp right upper abdominal pain, FHCS is sometimes confused with acute cholecystitis or pleurisy [6, 7]. A few cases have been reported in which patients with FHCS visited hospitals with complaints of pleuritic pain [2, 8]. However, the initial symptom is mainly abdominal pain. You *et al.* found that the chief complaints of 82 patients with FHCS were predominantly right upper quadrant (RUQ) pain in 58 cases (71%), lower abdominal pain in 5 cases (6.1%), right flank pain in 4 cases (4.9%), and pleuritic pain in only 1 case (1.2%) [2]. Woo *et al.* reported that the initial symptoms among 22 cases of FHCS were RUQ pain in 7 cases (32%), both RUQ pain and lower abdominal pain in 7 cases (32%), and solely lower abdominal pain in 6 cases (27%), with no cases of chest pain or pleuritic pain [3]. These studies indicate that the most frequent chief complaint is abdominal pain, especially in the RUQ, and that pleuritic pain is extremely rare.

In terms of differential diagnoses of RUQ pain, Middleton reported that FHCS was classified as a rare cause while differential diagnoses of RUQ pain were commonly biliary colic, cholecystitis, acute pancreatitis, acute appendicitis, alcoholic hepatitis, viral hepatitis, hepatic metastases, irritable bowel, and costochondritis (Thieme Medical Publishers. http://www.thieme.com/media/samples/pubid1184094957.pdf, accessed July, 2015). On the other hand, Kass *et al.* reported that differential diagnoses of pleuritic chest pain, as seen in our patient, included pulmonary embolism, myocardial infarction, pneumothorax, pleuritic, and pneumonia; FHCS was not listed as a cause of pleuritic chest pain [5]. These previous reports suggest that the initial symptom observed in the present case of FHCS was rare and difficult to diagnose because of the atypical chief complaint of chest pain combined with the lack of gynecological symptoms, for which she was first referred to a breast surgeon. Thus, when a patient complains of chest pain, it is difficult to make a diagnosis of FHCS because it is classified as an abdominal disease and because a complaint of chest pain is extremely rare. Even if a physician suspects abdominal disease, diagnosis is still difficult because basic diagnostic imaging of the abdomen, such as ultrasonography or plain CT, frequently show no abnormalities, as we experienced in the present case [3].

The mechanism underlying chest pain associated with FHCS is unclear. A case of FHCS that presented with right pleural effusion was previously reported, suggesting that perihepatitis and inflammation of the diaphragm might spread to the pleura and induce right pleural effusion [9]. Another hypothesis is that pain around the diaphragm can radiate up to just under the patient's right breast [10].

In our case, the patient did not demonstrate any signs or symptoms associated with PID, such as lower abdominal pain, vaginal discharge, or dysuria. A previous study reported that 60% of patients with FHCS complained of lower abdominal pain [3], of whom 41% presented with vaginal discharge [2]. The previous studies indicated that an absence of pelvic inflammatory symptoms cannot exclude a diagnosis of FHCS [2, 3]. Although a detailed sexual history of the patient was not obtained from the medical interview in our case, information about sexual activity has a critical role in the diagnosis of PID.

As a delay in the diagnosis and management of FHCS can cause serious and chronic complications, awareness of the condition is essential [11]. The present case suggests that, in a primary care setting, FHCS should be considered in young women with right-side chest pain even if they show no symptoms relevant to PID. Furthermore, specialists such as

cardiologists, pulmonologists, thoracic surgeons, and dermatologists may encounter such a patient as in the present case. Hence, their cooperation with general internists should be emphasized when treating patients who present with nonspecific initial symptoms. Also, general internists need to refer a patient to a gynecologist in an appropriate setting even if the patient complains of chest pain without any signs of gynecological disease, as in this case, because a cervical mucus sample plays an important role in the diagnosis of FHCS.

In conclusion, we find it necessary to consider FHCS, though rare, in the differential diagnosis of right-side chest pain in young women, as seen in this patient.

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