Biliary Taeniasis with Cholecystitis: An Unusual Case of *Taenia solium* Infection with a Literature Review

Hui-Jin Yu,¹† Chun-Seob Ahn,²† Sangeun Lim,¹ Jeong-Geun Kim,² Mi Sung Kim,³ Seoung Wan Chae,⁴ Joon-Sup Yeom,⁵ Eun-Jeong Joo,⁵ Woon-Mok Sohn,⁶ and Min-Jung Kwon^{1*}

¹Department of Laboratory Medicine, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea; ²Department of Molecular Parasitology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Suwon, Korea; ³Department of Radiology, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea; ⁴Department of Pathology, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea; ⁵Department of Internal Medicine, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea; ⁶Department of Parasitology and Tropical Medicine, Institute of Health Science, Gyeongsang National University College of Medicine, Jinju, Korea

Abstract. Taeniasis is a cosmopolitan helminthic disease caused by *Taenia* species, which included *Taenia solium*, *Taenia saginata*, and *Taenia asiatica*. These parasites typically infect the small intestine, but cases of aberrant migration have been reported. We treated a 70-year-old man who presented with vomiting and colicky abdominal pain. On physical examination, Murphy's sign was positive, and laboratory findings indicated severe inflammation. Computed tomography and magnetic resonance cholangiopancreatography revealed typical features of cholecystitis. An 82-cm-long, slender and degenerated, parasite-like organism was aspirated through a percutaneous transhepatic gallbladder drainage tube. After extensive washing of the organism, we detected yellowish-brown colored, spherical 37.9 × 33.8-μm-sized taenid eggs with thick transverse striations. Hematoxylin–eosin-stained worm sections also contained Taeniidae eggs. Polymerase chain reaction amplification of DNA extracted from the worm with species-specific cytochrome c1 (*cox1*) primer sets detected a *T. solium*-specific fragment. Because of sustained high fever combined with inflammatory signs, the patient underwent laparoscopic cholecystectomy and inflamed gallbladder removal. A histopathologic specimen demonstrated chronic reactive cholecystitis. The patient's fever and leukocytosis rapidly resolved after surgery. We experienced an uncommon case of biliary taeniasis representing cholecystitis caused by adult worm of *T. solium*.

INTRODUCTION

Human taeniasis is a disease complex caused by infection with *Taenia solium*, *Taenia saginata*, or *Taenia asiatica*. When humans ingest inadequately cooked pork or beef that is infected with the metacestodes of these tapeworms, the metacestodes develop into adults in the small intestine, where they may survive for long periods.¹ The disease occurs globally, but cases are more frequently reported from several Asian enclaves.² Taeniasis usually presents with vague, nonspecific gastrointestinal symptoms, which include abdominal discomfort, nausea, weakness, and weight loss.¹ In rare cases, the parasites cause effects similar to severe gastrointestinal or extraintestinal disease. Gastrointestinal diseases such as acute appendicitis or intestinal obstruction/perforation are associated with egg emboli, impacted worm segments, and strangled worm masses.^{3–6} Extraintestinal complications include involvement of the biliary system, pancreas, and liver. Several previous cases were mostly related to infections with *T. saginata* but not *T. solium*.^{2,5,7,8}

In this communication, we describe the case of a 70-year-old man with acalculous cholecystitis caused by adult *T. solium* impacted in the bile duct. The extreme rarity of the disease entity prompted us to report this case with a literature review.

MATERIALS AND METHODS

Laboratory examinations included analysis of complete blood cell count and serum levels of lactate dehydrogenase (LDH),

C-reactive protein (CRP), aspartate aminotransferase (AST), alanine aminotransferase (ALT), alkaline phosphatase (ALP), and γ-glutamyl transpeptidase (γ-GTP). Radiographic examinations using contrast-enhanced computed tomography (CT) and magnetic resonance cholangiopancreatography (MRCP) were conducted to evaluate the patient's suspected cholangitis.

A parasite-like creature drained through percutaneous transhepatic gallbladder drainage (PTGBD) was identified by morphologic and genetic analyses. We thoroughly washed the organism with cold physiological saline more than 10 times. The washing fluid was centrifuged at 1,000 × *g* for 5 minutes, after which the sediments were subjected to microscopic examination. The posterior part of the worm was fixed and processed for histological examination. Acetocarmine-stained worm specimen and hematoxylin–eosin (H&E)-stained worm sections were microscopically observed. For genetic assessments, we amplified the worm's cytochrome c1 (*cox1*) gene by polymerase chain reaction (PCR). We retrieved genomic sequences of *T. solium* *cox1* gene (TsM_000113400) from GeneDB (<http://www.genedb.org/Homepage/Tsolium>) and designed a specific primer set (nucleotide positions between 18899 and 19955 in TsM_scaffold_01048 [17888–20955]): forward 5'-GCGTGGCTGTTGGTTCC-3' and reverse 5'-CTACA GTAGACACCACTT-3'. We extracted genomic DNA from the parasite together with that from known *T. solium*, *T. saginata*, and *T. asiatica* samples⁹ using a commercially available kit (QIAamp DNA mini kit; Qiagen, Valencia, CA). The PCR cycling parameters were 5 minutes at 95°C, 35 cycles of 1 minute at 95°C, 45 seconds at 50°C, 1 minute at 72°C, and a final extension of 5 minutes at 72°C. This study was approved by the institutional review board of Kangbuk Samsung Hospital, Sungkyunkwan University (KBSMC 2018-02-044).

We surveyed literatures in the publicly available domains of the National Center for Biotechnology Information (PubMed)

* Address correspondence to Min-Jung Kwon, Department of Laboratory Medicine, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, 29 Saemunan-ro, Jongno-gu, Seoul 03181, Korea. E-mail: mjkkmd@gmail.com

† These two authors contributed equally to this work.

and Google Scholar using the term “*Taenia*” in conjunction with “cholecystitis”, “gallbladder”, “bile duct”, and “biliary tract”. After relevant publications were retrieved, we also reviewed the references cited by these publications to identify additional cases.

REPORT OF A CASE

A 70-year-old male patient visited a local clinic with chief complaints of recurrent vomiting and abdominal colicky pain. Ultrasonographic examination suggested acute cholecystitis. He was transferred to our institute. His vital signs were stable but we noted a fever (38.0°C). On physical examination, Murphy's sign was positive. Complete blood count revealed leukocytosis ($20.6 \times 10^9/L$), with a predominance of neutrophils (84%). Elevated serum levels of LDH (275 IU/L) and CRP (26.84 mg/dL) indicated a severe inflammatory status. Elevated serum level of fasting glucose (169 mg/dL) and 7.6% HbA1c were first noticed, and then diagnosed as diabetes mellitus in our hospital. Results of AST, ALT, ALP, and γ -GTP levels were normal. A preoperative, transverse CT image exhibited focal disruptions of the enhancing mucosa (arrows) of the thickened gallbladder wall, suggesting agangrenous cholecystitis (Figure 1A). An axial T2-weighted image revealed pericholecystic edema and inflammation (Figure 1B). We recognized hypointense linear structures (arrows) within the intra- and extrahepatic bile ducts on a single-shot MRCP image (Figure 1C and D). The patient did not recognize any significant medical history of prior hepatobiliary diseases, parasitic infections, and digestive problems, including loss of appetite, weight loss, upset stomach, and abdominal colic pain. He also denied passing any parasite-like organism

through the anus or in the feces. He assumed that he usually did not eat an uncooked food. The patient was immediately prescribed with ceftriaxone and metronidazole.

Percutaneous transhepatic gallbladder drainage was carried out on the third day of admission. During irrigation of the inserted catheter, a bile-tinged, parasite-like organism was aspirated (Figure 2A). The creature had a long, slender, glistening, opal-colored body measuring up to 82 cm (3–5 mm in width) with a single dark-brown line running along the entire body. Two knots were observed. The anterior and posterior ends of the organism were not found, but proglottid-like structures, which showed very little movement, were recognized under a dissecting microscope (arrowheads). We extensively washed the worm with ice-chilled physiological saline. Washing fluid was briefly centrifuged and sediments were observed with microscopy. We identified Taeniidae eggs whose sizes ranged from $37.5 \times 38.8 \times 32.5$ to $35.0 \mu m$ (mean: $37.9 \times 33.8 \mu m$). The embryophore was covered with thick transverse striations. Hooklets were found during fine adjustment of the microscope (Figure 2B). The posterior part of the worm, which was fixed and stained with acetocarmine, showed highly degenerated proglottid-like structures. However, internal organs characteristic to *Taenia* were not recognized, except for numerous brown dots that might correspond to a dark-brown line running along the body (asterisk, Figure 2C).

We prepared H&E-stained worm sections to observe possible structures/organs specific to *Taenia* worm (Figure 2D). Highly degenerated, laterally positioned reproductive systems were recognized, in which some *Taenia* eggs were observable (arrows, Figure 2E and F). To further identify the parasite species, we performed PCR using DNA extracted from the

FIGURE 1. Computed tomography and magnetic resonance cholangiopancreatography (MRCP) images. (A) Focal disruptions of the enhancing mucosa (arrows) of the thickened gallbladder wall suggest agangrenous cholecystitis. (B) An axial T2-weighted image revealed pericholecystic edema and inflammation. (C and D) Hypointense linear structures (arrows) within the intra- and extrahepatic bile ducts are noticed on a single-shot MRCP image.

FIGURE 2. Morphological and molecular identification of *Taenia solium* extracted from the patient. (A) A broken parasite-like creature was drained from the gallbladder through a T-tube. The opal-colored, glistening organism had a long narrow body (82 cm) and was knotted. Bar = 5 cm. (B) Taenid eggs observed in washing fluid. The eggs are dark-brown, spherical, and contained embryophore covered with thick transverse striation. They ranged in size from 37.5 to 38.8×32.5 to 35.0 μm (mean: 37.9×33.8 μm). Bar = 10 μm . (C) The distal portion of the parasite was cut and subjected to acetocarmine staining. A highly degenerated proglottid-like architecture was observed. Bar = 2.5 mm. (D) Cross-sectioned worm specimen stained with hematoxylin–eosin reveals a highly degenerated reproductive system, which is located in the lateral portion of the proglottid. (E and F) Highlighted view of the reproductive system marked by the box in D and E. Taenid eggs are observed (arrows). Bar in F denotes 50 μm . (G) Amplification of the *T. solium* cytochrome c oxidase 1 (*cox1*) gene by polymerase chain reaction (PCR). DNA samples extracted from known *T. solium*, *Taenia saginata*, and *Taenia asiatica* samples together with samples from the patient were subjected to PCR using a primer set specific to the *T. solium cox1* gene. M, 1-kb ladder. This figure appears in color at www.ajtmh.org.

worm. A 1,057-base pair-long segment was specifically detected with a *T. solium* sample in this study. *Taenia saginata* and *T. asiatica* DNAs did not produce amplicons (Figure 2G). We also conducted PCR with specific primer sets for *T. saginata* and *T. asiatica cox1* genes, but did not observe any specific band (data not shown).

His gastrointestinal symptoms subsided after removing the worm from the bile duct. The patient was further treated with a single dose of praziquantel (10 mg/kg body weight). Because of sustained fever combined with inflammatory signs, we removed the patient's inflamed gallbladder through laparoscopic cholecystectomy on the ninth day of admission (Figure 3A). An H&E-stained histopathologic specimen demonstrated characteristic features of chronic reactive cholecystitis, including regenerating mucosal glands and multifocal ulcerated lesions with a fibrinoid crust and foamy histiocytic deposition. Lymphoplasm and eosinophilic infiltration were also noted, but residual parasitic worms or eggs were not observed (Figure 3B). The patient's body temperature, leukocyte counts, and CRP level returned to normal ranges after

cholecystectomy and medical treatment. He demonstrated clinical improvement and had recovered to good condition 1 month after the laparoscopic cholecystectomy.

DISCUSSION

A nationwide survey in Korea showed that the *Taenia* egg-positive rate ranged from 0.01% to 0.04% during the past two decades, suggesting that taeniasis is very rare in Korea.¹⁰ Most intestinal taeniasis cases are asymptomatic or present with nonspecific, mild symptoms. Extraintestinal taeniasis caused by tapeworms is a rare condition even in endemic areas. We reviewed the literatures to search for cases of hepato-pancreato-biliary taeniasis. A total of 15 cases excluding our present case have been reported (Table 1). Seven cases were from Europe: three from France, two from Turkey, and one each from Sweden and Ireland. The remaining eight cases were from Asia and the Americas: two from India and one each from Georgia, Saudi Arabia, Taiwan, Korea, the United States, and Uruguay. The male-to-female ratio was

FIGURE 3. Gross and histopathologic specimens of the gallbladder isolated from the patient. (A) The resected gallbladder shows chronic cholecystitis. Bar = 2 cm. (B) Hematoxylin-eosin-stained histopathologic specimen reveals regenerating mucosal glands, multifocal ulcerated lesions, and fibrinoid crust. Infiltrations of lymphoplasmic cells and eosinophils are observed. Fatty necrosis and foamy histiocytic collections under the ulcerated lesions are also observed. This figure appears in color at www.ajtmh.org.

2:3, with an average age of 60.0 years for males and 50.6 years for females. All previously reported extraintestinal cases were caused by *T. saginata* or possibly by *T. asiatica* according to morphological identification (Table 1). Our case is presumed to be the first case of infection by *T. solium* involving the biliary system.

The clinical manifestations and imaging findings of our patient were compatible with acalculous cholecystitis. To relieve his acute and debilitating symptoms, we inserted a PTGBD, and a parasite-like organism was incidentally drained during irrigation. The creature initially appeared to be a nematode because it had a long, slender body with a tough and glistening surface. However, it was too long to be a nematode and was somewhat flattened rather than round. The worm was knotted and both ends were broken (Figure 2A). These observations suggested that the worm might be a cestode, because the elastic and collagen-rich nematode cuticle is highly flexible and is not easily broken or knotted.¹¹ Moreover, we found eggs morphologically characteristic to taenid tapeworms (Figure 2B). We prepared worm specimens for further identification but we could not observe any organ/structure specific to *Taenia* worm (Figure 2C). We examined the H&E-stained sectioned worm (Figure 2D) and found highly degenerated reproductive systems that contained *Taenia* eggs (Figure 2E and F). The taenid reproductive system is usually positioned in the central part of each proglottid. However, we observed laterally located, highly degenerated reproductive organs.

TABLE 1
Reported cases of hepato-pancreato-biliary taeniasis

No	Year	Place	Age/sex	Affected organs	Reference
1	1926	United States	74/M	Gallbladder	15
2	1949	Sweden	51/F	Bile duct	16
3	1954	Uruguay	44/M	Gallbladder	17
4	1957	France	69/F	Liver	7
5	1960	Ireland	69/F	Bile duct	18
6	1968	Georgia	55/F	Gallbladder	19
7	1980	France	73/M	Pancreatic duct	20
8	1981	Korea	77/M	Gallbladder	21
9	1998	France	60/M	Gallbladder	22
10	1999	Turkey	35/F	Gallbladder	23
11	2005	Taiwan	52/F	Pancreatic duct	2
12	2008	India	26/F	Gallbladder	24
13	2012	India	32/M	Gallbladder	4
14	2012	Turkey	58/F	Gallbladder	5
15	2014	Saudi Arabia	40/F	Pancreatic duct	8
16	2018	Korea	70/M	Gallbladder	Present case

F = female; M = male.

This result might reflect that the reproductive system might be injured in its central position through loss of supporting parenchymal matrix during degenerative changes. It might be located on the dependent position. These collective data indicated that the parasite was *Taenia* spp. However, human-infecting *Taenia* parasites have morphologically indistinguishable eggs.¹² We amplified a *cox1* gene for species identification, since mitochondrial *cox1* genes are a useful biomarker for interspecies differentiation of human-infecting *Taenia* spp.⁹ We were able to amplify a *cox1* gene specific to *T. solium* and concluded that the parasite triggering cholecystitis in our patient was *T. solium* (Figure 2G).

Several helminth parasites can ectopically migrate to the biliary system. The most frequent cases involve *Ascaris* because this worm is highly motile in the intestine.¹³ Among taenid parasites, *T. saginata* and *T. asiatica* also have active motility in the intestine.¹⁴ These parasites are occasionally found in the biliary tract, including the gallbladder and pancreas (Table 1). Conversely, *T. solium* usually shows only passive movement.¹² The poor motility of *T. solium* might explain the rare incidence of extraintestinal migration by this species. The worm extracted from our patient showed highly degenerative changes. Histopathologic examination of the gallbladder specimen also revealed chronic reactive cholecystitis (Figure 3A and B). These results suggest strongly that the parasite may have silently resided in the biliary tract and/or gallbladder for an extended period of time due mainly to poor motility. Unknown exacerbating factors then abruptly provoked acute symptoms that manifested as acalculous cholecystitis.

In conclusion, we report a *T. solium* infection of the gallbladder invoking reactive cholecystitis in a Korean male. This report has worth in the symptomatic infection caused by adult worm, not the larval form of *Taenia*. Although such cases are exceptional, the possibility of *T. saginata*, *T. asiatica*, or *T. solium* presence in the biliary duct should be kept in mind during clinical practice, especially in endemic areas.

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Authors' addresses: Hui-Jin Yu, Sangeun Lim, and Min-Jung Kwon, Department of Laboratory Medicine, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea, E-mails:

hjhj0821@gmail.com, sangeun87.lim@samsung.com, and mjkkmd@gmail.com. Chun-Seob Ahn and Jeong-Geun Kim, Department of Molecular Parasitology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Suwon, Korea, E-mails: feg123@naver.com and chakangni@hanmail.net. Mi Sung Kim, Department of Radiology, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea, E-mail: misung70@gmail.com. Seoung Wan Chae, Department of Pathology, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea, E-mail: swan.chae@samsung.com. Joon-Sup Yeom and Eun-Jeong Joo, Department of Internal Medicine, Kangbuk Samsung Hospital, Sungkyunkwan University School of Medicine, Seoul, Korea, E-mails: joonsup.yeom@gmail.com and eunjeong.joo@samsung.com. Woon-Mok Sohn, Department of Parasitology and Tropical Medicine, Institute of Health Science, Gyeongsang National University College of Medicine, Jinju, Korea, E-mail: wmsohn@gnu.ac.kr.

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