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Spontaneous cholecystopleural fistula leading to biliothorax and sepsis in a cat

Gretchen M VanDeventer[1,2] and Benoît Y Cuq[3]

Abstract

Case summary A 13-year-old spayed female domestic shorthair cat presented with pleural effusion and suspected triaditis. Intake vitals and leukocytosis were consistent with a diagnosis of systemic inflammatory response syndrome. Biochemical analysis confirmed a pleural fluid-to-serum bile ratio consistent with a diagnosis of biliothorax. Abdominal ultrasound failed to identify a definitive gall bladder but noted a hypoechoic tubular structure ventral to the liver and contacting the diaphragm. Thoracic ultrasound identified a hyperechoic structure contacting the diaphragm at the same location. Thoracoabdominal CT scan identified a fluid-dense tubular structure extending from ventral to the liver, through a diaphragmatic defect and directly communicating with the pleural space, suspected to be an abnormal gall bladder. The cat was humanely euthanized, and post-mortem analysis confirmed a cholecystopleural fistula arising from the gall bladder with multifocal abscesses, mixed inflammatory hepatic infiltrates and small-cell gastrointestinal lymphoma. Culture of the abscess isolated *Parabacteroides merdae*, meeting the reported feline criteria for sepsis.

Relevance and novel information To our knowledge, spontaneous cholecystopleural fistula formation leading to biliothorax and sepsis has not been previously reported in the cat. This case highlights a novel sequela of gall bladder disease in this species, and biliothorax should be a differential diagnosis for pleural effusion in cats with evidence of cholecystitis or triaditis.

Keywords: Biliothorax; cholecystopleural fistula; triaditis; sepsis; systemic inflammatory response syndrome; SIRS

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Case description

A 13-year-old 3.0 kg (6.6 lb) spayed female domestic shorthair cat presented to the Companion Animal Hospital at the Ontario Veterinary College for evaluation of pleural effusion and suspected pancreatitis. Fourteen days prior, the referring veterinarian assessed the cat for acute respiratory distress, lethargy and anorexia. Thoracic radiographs at that time were unremarkable (Figure 1). Serum biochemistry analysis revealed a moderately elevated alanine aminotransferase concentration (283 IU/l; reference interval [RI] 27–158 IU/l), mild total hyperbilirubinemia (7.7 µmol/l; RI 0–5.13 µmol/l) and an elevated feline pancreas-specific lipase (Spec fPL-Feline test; IDEXX Laboratories) (15.0 µg/l; RI 0–3.5 µg/l). A complete blood count (CBC) was unremarkable. The cat was treated as an outpatient with amoxicillin–clavulanic acid, buprenorphine, subcutaneous fluid therapy and syringe feeding, and the respiratory signs initially improved within 24 h of presentation.

At a recheck evaluation performed 7 days later, a new hyperthermia of 40°C was identified. Serum biochemistry analysis revealed resolution of the previously abnor-

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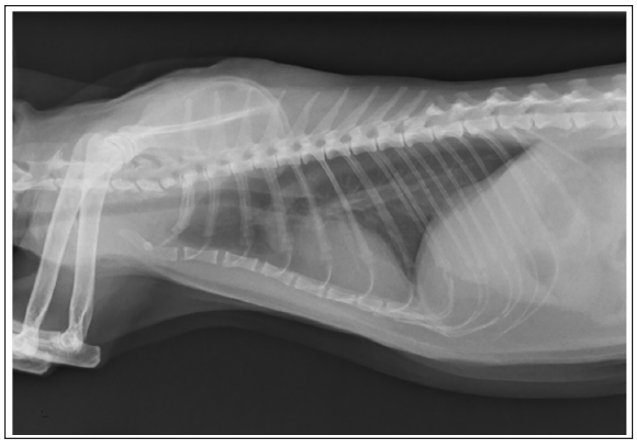

Figure 1 Right lateral thoracic radiograph depicting normal thorax at initial presentation to the primary veterinarian despite acute dyspnea

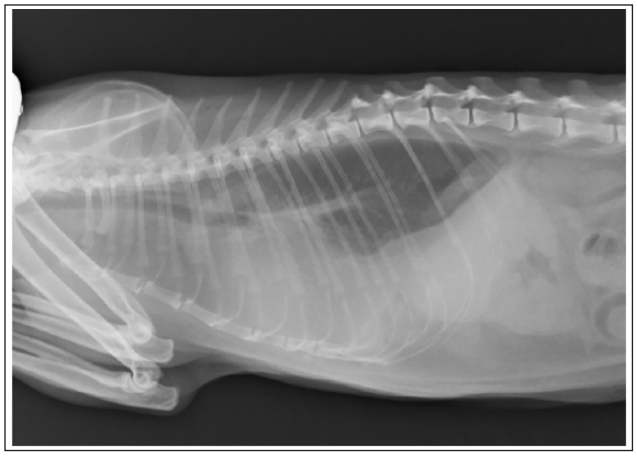

Figure 2 Right lateral thoracic radiograph performed 14 days after initial presentation to the primary veterinarian depicting moderate caudoventrally located pleural effusion

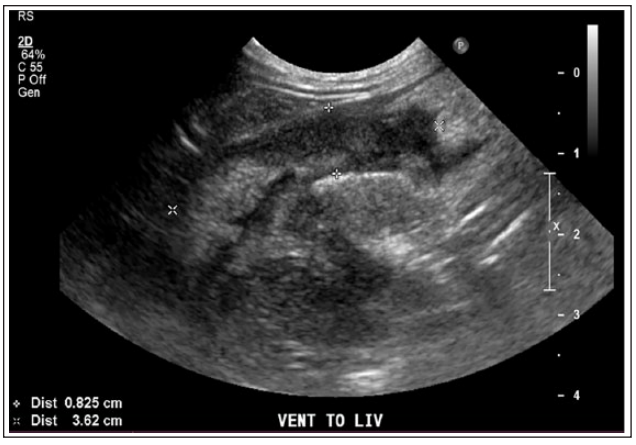

Figure 3 Abdominal ultrasound image depicting tubular structure ventral to and, questionably, arising from the right liver

mal parameters, but a mature neutrophilia (23.0 × 10^9/l; RI 2.6–15.2 × 10^9/l) had arisen. The cat began eating small amounts voluntarily and no further respiratory distress had occurred. The medications were continued for five more days, until the cat again became anorexic and lethargic. At that time, a urinalysis revealed bilirubinuria and serum biochemistry analysis identified moderate total hyperbilirubinemia (10.0 µmol/l). A CBC revealed a progressive mature neutrophilia (30.0 × 10^9/l). The following day the cat became severely dyspneic, and thoracic radiographs revealed moderate right-sided pleural effusion (Figure 2). Thoracocentesis yielded a thick yellow–orange fluid, and in-house cytological evaluation of the fluid revealed leukocytes without evident bacteria. The dyspnea improved following the thoracocentesis procedure. Overnight hospitalization with buprenorphine, amoxicillin–clavulanic acid and furosemide was pursued, and the cat was referred the following day.

On physical examination, the cat was in poor body condition (body condition score 3/9), depressed, weakly ambulatory and approximately 10% dehydrated. Normothermia (38.6°C; 101.5°F), tachycardia (280 beats per min) and mild tachypnea (52 breaths per min) were noted, and lung sounds were decreased on the right side. A grade II/VI left systolic heart murmur was auscultated. No other specific abnormalities were identified on physical examination. Point-of-care ultrasound evaluation identified mild right-sided hypoechoic pleural effusion, scant hypoechoic pericardial effusion and no abdominal effusion; thoracocentesis was not performed. Blood gas and electrolyte analysis revealed a mild hyperglycemia (11.8 mmol/l; RI 4.4–7.7 mmol/l), hypochloremia (97 mmol/l; RI 114–123 mmol/l) and hypokalemia (3.4 mmol/l; RI 3.6–5.2 mmol/l). The patient fulfilled the criteria for the systemic inflammatory response syndrome (SIRS) based on the tachycardia, tachypnea and leukocytosis identified on recent CBC.[1] Intravenous fluid therapy was initiated using 12 ml/h of a balanced crystalloid solution (Plasmalyte-A; Baxter Canada) with 20 mEq/l potassium chloride supplementation.

Cytologic evaluation of the pleural fluid sampled by the primary veterinarian the evening prior revealed marked non-septic suppurative inflammation with frequent amorphous-to-crystalline yellow material consistent with bilirubin crystals. Biochemical analysis indicated that the fluid was consistent with bile (pleural fluid bilirubin: 286 µmol/l; peripheral blood bilirubin: 10 µmol/l). The fluid-to-serum bilirubin ratio (>28) was consistent with a diagnosis of biliothorax.

Abdominal and thoracic ultrasound examinations were performed. The gall bladder could not be readily identified on abdominal ultrasonography. A small hypoechoic structure with irregular margins that contacted the diaphragm was identified, ventral to the right side of the liver (Figure 3). This structure contacted the liver and

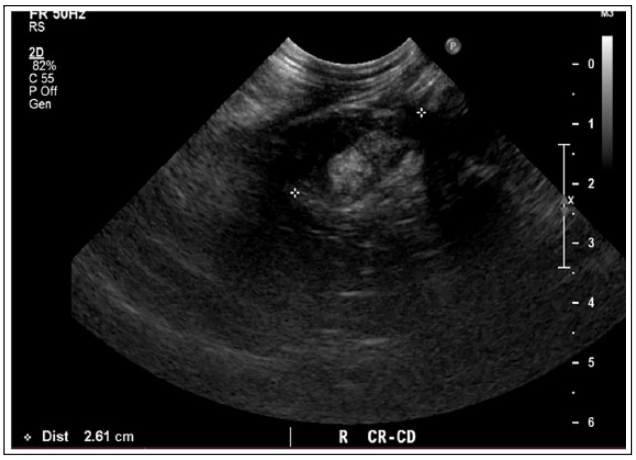

Figure 4 Thoracic ultrasound image depicting a hyperechoic structure in right caudal thorax, in contact with the diaphragm. There is moderate pleural effusion ventral to the structure

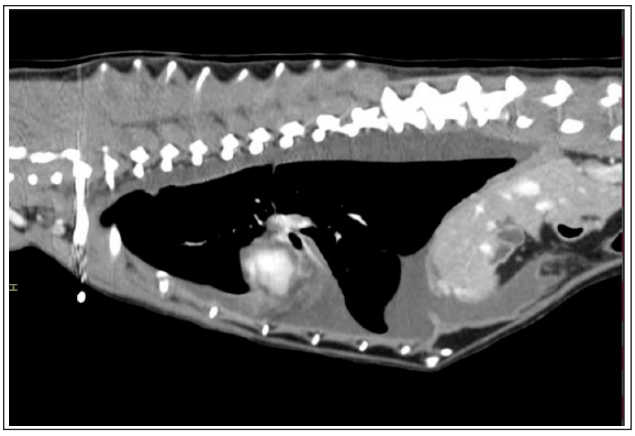

Figure 5 Sagittal view of thoracoabdominal CT scan depicting open communication between the fluid-filled abdominal structure and the thorax via a defect in the diaphragm

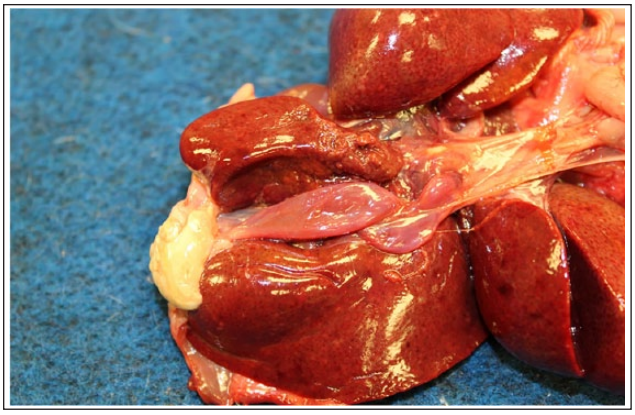

Figure 6 Necropsy image. Visceral surface of the liver with abscessed tissue at left connecting deflated gall bladder and diaphragm

in some areas appeared to be arising from it. Differential diagnoses for the structure included: a neoplastic mass arising from the liver, gall bladder or peritoneal space; a displaced and abnormal gall bladder secondary to neoplasia, cholecystitis or rupture; or a neoplastic or torsed liver lobe.

Thoracic ultrasonography identified mild pericardial and moderate pleural effusion, and a mild thickening of the parietal pleura. A hyperechoic structure was identified in the right caudal thorax in contact with the diaphragm, symmetrical to the hypoechoic abdominal structure (Figure 4). No specific diaphragmatic defects were appreciated, but an abnormal communication between the thorax and abdomen could not be excluded via either an anatomical defect such as a peritoneopericardial diaphragmatic hernia or a de novo communication such as that due to transdiaphragmatic invasion of an aggressive liver or biliary lesion.

Thoracic and abdominal CT scans were then performed under general anesthesia to further characterize the abnormal structures identified on ultrasonography, and to investigate possible communication between the thorax and abdomen. This revealed a fluid-dense tubular structure extending from ventral to the liver, through the diaphragm, into the thorax, and directly communicating with the pleural space (Figure 5). This was suspected to be an abnormal gall bladder. There was no evidence of a peritoneopericardial diaphragmatic hernia. Filling defects were also detected in the caudal lobar pulmonary arteries, consistent with pulmonary thromboemboli, and moderate sternal lymphadenopathy was identified, suspected to be due to reactive lymphadenitis or metastatic neoplasia. The patient was unstable during the general anesthetic event, exhibiting marked bradycardia and hypotension despite appropriate intervention and support.

An aggressive neoplastic or infectious lesion of the biliary tree with focal diaphragm destruction was suspected. Although a traumatic gall bladder herniation through a diaphragmatic tear with subsequent strangulation and necrosis could not be excluded, it was considered less likely based on the patient's history. As the cat was unstable under anesthesia and an aggressive lesion was suspected, the owner elected humane euthanasia rather than pursuing further intervention.

A necropsy was performed, which identified a cholecystopleural fistula arising from the apex of the gall bladder (Figure 6) with multifocal abscesses and cellular necrosis, and severe acute fibrinous pleuritis. Mixed neutrophilic, lymphoplasmacytic and mononuclear inflammatory infiltrates were identified in the liver, as were changes consistent with hepatic lipidosis. No abdominal effusion was identified, but histologic evaluation of the spleen identified plump mesothelial cells lining the capsular surface, consistent with chronic bile

irritation. An incidental thyroid adenoma and small-cell intestinal lymphoma were also identified. A sample of the abscess exudate was submitted for bacterial culture, which isolated *Parabacteroides merdae*.

Discussion

Biliothorax, also termed bilothorax or bile pleuritis, is a rare condition in which bile accumulates within the pleural space. Although gross appearance of the pleural effusion can raise suspicion for this condition, definitive diagnosis is based on a ratio of the pleural fluid-to-serum bilirubin concentration that is >1.[2,3] In the present case, the fluid was a thick yellow–orange color, and the ratio was >28 (pleural fluid bilirubin: 286 μmol/l; peripheral blood bilirubin: 10 μmol/l), confirming the diagnosis of biliothorax.

Isolated case reports of biliothorax have previously been described in five dogs and three cats, all of which were due to traumatic or iatrogenic injuries.[4–11] The most commonly reported cause for this condition has been diaphragmatic and biliary tree laceration secondary to gunshot wounds,[4–7] while one cat developed the condition after bite wounds led to diaphragmatic injury and gall bladder herniation.[8] A cat was also reported to develop iatrogenic biliothorax as a complication of a thoracostomy tube placement.[9] Two further dogs developed biliothorax in the absence of overt diaphragmatic injury, one after vehicular trauma led to rupture of the extrahepatic biliary tract,[10] and the other after a cholecystectomy surgery.[11] Of these cases, concurrent bile peritonitis was identified in four cases secondary to direct trauma to the biliary tree.[4,6,8,10] None of these cases, however, involved a biliothorax associated with development of a spontaneous cholecystopleural fistula secondary to cholecystitis.

Cholecystitis is a relatively common condition in cats, and has been extensively reviewed.[12,13] Although cholecystitis can occur as a unique pathology, in this case the patient was suspected based on serum biochemistry and post-mortem histopathologic changes to have developed triaditis: concurrent inflammation of the pancreas, liver and small intestine. Triaditis can develop from primary pancreatitis leading to intestinal inflammation and reactive cholangiohepatitis due to the close proximity of the common bile duct and pancreas in the duodenum, or from chronic gastrointestinal inflammation leading to reactive pancreatitis and cholangiohepatitis.[14] Regardless of the inciting cause, inflammation in the region of the pancreatic and common bile ducts can predispose animals to bacterial infection of the biliary tree via both retrograde migration of jejunal bacteria and via translocation from hepato-biliary-enteric circulation.[12]

In this case chronic intestinal inflammation secondary to small-cell lymphoma was identified on post-mortem examination, which may have been an inciting cause for the pancreatitis and cholangiohepatitis. Alternatively, an episode of acute pancreatitis may have developed in the face of small-cell gastrointestinal lymphoma with subsequent cholangiohepatitis. While triaditis is well described, progression to a cholecystopleural fistula is not well documented.

None of the previously reported cases of biliothorax in veterinary patients isolated a positive bacterial culture. Although ante-mortem cytological evaluation of the pleural fluid identified suppurative but not septic inflammation, post-mortem bacterial culture of the abscess isolated *P merdae*.

The *Parabacteroides* species were recently divided from the *Bacteroides* species,[15] and these groups constitute the most common anaerobic bacteria isolated from hepatic and biliary bacterial cultures in cats, dogs and humans,[16–18] and are also among the most common bacteria isolated from feline pyothorax.[19] Although the *Parabacteroides* species have not been identified as specific pathogens in cats, the closely related *Bacteroides* species have been implicated in a variety of health conditions in humans, including septic peritonitis, meningitis and endocarditis, and have evolved strategies for bile tolerance.[20] Although *P merdae* was identified on culture of the abscess, it is unknown whether it was acting as a pathogen alone or in combination with another bacterial species that was inhibited or killed by the prior antibiotic therapy, or alternatively was an incidental colonizer, with the abscess formed by a separate species that was inhibited or killed by the antibiotics.

It has been well established in veterinary species that septic bile peritonitis has a significantly higher mortality rate than sterile bile peritonitis, with 27% and 100% survival, respectively.[21] Given the rarity of the condition, no similar studies are available comparing septic biliothorax and aseptic biliothorax in animals; however, it is reasonable to anticipate a similar pattern.

Diagnosis of sepsis involves meeting the criteria for SIRS with concurrent positive bacterial infection based on cytologic, histologic, microbial or serologic tests.[1] The patient in this case met the criteria for SIRS based on leukocytosis and on tachycardia and tachypnea that did not respond to early fluid resuscitation, and also fulfilled the criteria for sepsis based on the positive bacterial culture obtained from the post-mortem abscess sample. Sepsis has been associated with substantial morbidity to various organ systems, as well as mortality rates ranging from 20–79% in cats.[1,22–25] Hypochloremia, which was documented in this case, has been associated with a negative prognosis in cats with sepsis.[26] Cats also appear uniquely sensitive to pulmonary damage secondary to SIRS and sepsis compared with dogs, as the lungs are a major source of bacterial clearance and inflammatory reaction in this species.[27,28] The patient in this report did have suspicion for pulmonary thromboemboli on CT scan, which is a

reported complication of SIRS and sepsis and corroborates our suspicion of sepsis in this case.[1] This likely contributed to the instability exhibited under general anesthesia, and was considered a poor prognostic indicator.

Biliothorax has also been described uncommonly in human medicine, generally secondary to the development of a biliopleural fistula, although it has also been rarely reported as a complication of bile peritonitis in the face of an intact diaphragm.[2,3] Causes for fistula formation include iatrogenic injury secondary to diagnostic and therapeutic interventions,[29,30] thoracoabdominal trauma,[31] parasitic liver disease,[32–34] and suppurative and non-suppurative biliary tract obstructions.[2,35–37] Spontaneous cholecystopleural fistulae most commonly develop in human patients due to suppurative biliary tract obstruction or chronic cholecystitis secondary to gallstones.[35,36,38] Successful management has been reported following prompt medical and surgical therapy,[35,39] but prognosis is poor if diagnosis and treatment are delayed, as biliothorax can lead to severe manifestations of a systemic inflammatory response and death.[38,39]

The prognosis for biliothorax in veterinary patients has been reported to be excellent, with all eight previously described cases surviving to discharge, although case selection bias may have influenced this reported outcome. Two of these cases were managed with medical therapy alone,[10,11] while six underwent surgical intervention.[4–9] It is ultimately unknown whether medical or surgical therapy would have been successful in the present case; however, multiple factors contributed to a potentially worse prognosis than that noted in previous cases, including the development of sepsis and associated complications. Prolonged empiric therapy and delayed diagnosis of the gall bladder abscess that led to the cholecystopleural fistula and biliothorax likely also contributed to the patient's worse prognosis.

Conclusions

This case highlights a novel sequela of gall bladder disease in the cat: biliothorax secondary to a spontaneous cholecystopleural fistula. Given the importance of early diagnosis and intervention in these cases, biliothorax should be a differential diagnosis for development of pleural effusion in patients with evidence of cholecystitis or triaditis. Delayed intervention may increase the risk of development of SIRS or sepsis, which may negatively affect the prognosis. Although treatment was not pursued in this case, extrapolation from human medicine suggests that surgical debridement, cholecystectomy and closure of the defect would be indicated.

Conflict of interest The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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