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CASE REPORT
Biliothorax in a Siamese cat

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A 2-year-old male neutered cat presented for further investigation of biliothorax. The cat was initially treated for pyothorax, including bilateral chest drains for lavage of the pleural space. Five days later, the pleural effusion turned clear-yellow and had a bilirubin concentration of 427 μmol/l compared to the serum bilirubin concentration of 15 μmol/l. Exploratory surgery revealed a 2 mm tear in the diaphragm, with a corresponding 2 mm defect in the diaphragmatic surface of the gall bladder, creating a fistula between the gall bladder and the pleural cavity. The defects were repaired routinely and the cat made a full recovery. It was suspected that the tears had been created at the time of the thoracostomy tube placement. Biliothorax has not been described before in a cat, and appears to be a rare complication following thoracostomy tube placement.

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A 2-year-old, male neutered, Siamese cat was presented to the Small Animal Teaching Hospital (SATH), University of Liverpool, for investigation of a biliary pleural effusion. Eight days earlier, the referring veterinarian had assessed the cat for acute respiratory distress. Cytological evaluation of pleural fluid revealed degenerate neutrophils with intracellular bacteria, and a diagnosis of pyothorax was made. However, bacterial culture of the fluid was negative. Thoracostomy drainage tubes were placed, bilaterally, to facilitate drainage and intermittent pleural lavage with 0.9% saline. In addition, intravenous broad-spectrum antibacterial therapy was administered, using a combination of amoxicillin–clavulanic acid and metronidazole. Pleural fluid and serum were submitted for feline *Coronavirus* species antibody measurement, and results were negative. After 5 days of therapy, the nature of the fluid changed from turbid and opaque to clear-yellow. The drains continued to yield 10–20 ml/day, although the dyspnoea resolved with this management. However, the cat continued to be depressed, lethargic and anorexic. Analysis of the pleural fluid revealed a total bilirubin concentration of 427 μmol/l, compared to the serum total bilirubin concentration of 15 μmol/l (reference interval 0–15 μmol/l) and a total protein content of 2.9 g/dl (reference interval <2.5 g/dl). A diagnosis of biliothorax was made and the cat was referred at this stage.

On physical examination, the cat was in poor body condition (body condition score (BCS) 3/9), depressed, mildly tachypnoeic and approximately 7% dehydrated. Both thoracostomy tubes remained in place, with a crusted discharge present around their skin wounds. Haematology showed a left shift neutrophilia and biochemistry was consistent with dehydration but also showed two-fold elevation in alkaline phosphatase and three-fold elevation of alanine transferase (Table 1). Intravenous fluid therapy was initiated using 12 ml/h of a crystalloid solution (Aquapharm; Hartmann's solution, Animal Care, UK) and the antibacterial therapy was continued with amoxicillin–clavulanic acid (Augmentin; GlaxoSmithKline, UK) at 20 mg/kg, IV, q 8 h and metronidazole (Metronidazole; Baxter Health Care, UK) at 10 mg/kg, IV, q 12 h. The pleural space was drained twice daily via the thoracostomy tubes already placed by the referring veterinarian and the amount recorded. Thoracic radiographs were obtained under sedation to evaluate the thoracostomy tube positioning and further investigate the effusion (Fig 1a and b). Thoracic and abdominal ultrasound examinations were performed. Thoracic ultrasonography

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Table 1. Clinicopathological parameters on presentation. Values outwith the reference interval are in bold. The cat shows a left shift neutrophilia, mild pre-renal azotaemia consistent with dehydration. Other causes of the raised urea and normal creatinine could be a recent protein meal or gastrointestinal haemorrhage. The mild electrolyte disturbances were not considered to be of clinical significance.

Parameter	Measurement	Reference interval
Red blood cell count ($\times10^{12}$/l)	6.65	5–11
Haemoglobin (g/dl)	12.2	8–11
Haematocrit (l/l)	0.32	0.26–0.46
MCV (fl)	48	37–49
White cell count ($\times10^{9}$/l)	**30.4**	5.5–19.5
Mature neutrophils ($\times10^{9}$/l)	**26.5**	2.5–12.5
Band neutrophils ($\times10^{9}$/l)	**0.59**	0–0.3
Lymphocytes ($\times10^{9}$/l)	2.36	1.5–7.0
Monocytes ($\times10^{9}$/l)	0.44	0–0.85
Eosinophils ($\times10^{9}$/l)	0.44	0.1–1.5
Basophils ($\times10^{9}$/l)	<0.01	0–1.0
Total protein (g/l)	67	55–78
Albumin (g/l)	22	20–30
Globulin (g/l)	45	26–51
Sodium (mmol/l)	156	145–156
Potassium (mmol/l)	**5.7**	3.8–5.3
Chloride (mmol/l)	**113**	117–140
Total calcium (mmol/l)	2.35	2.1–2.6
Inorganic phosphorus (mmol/l)	1.69	1.1–2.3
Urea (mmol/l)	**10.4**	2.5–7.5
Creatinine (μmol/l)	88	40–120
Cholesterol (mmol/l)	2.3	1.9–3.9
Alkaline phosphatase (IU/l)	91	0–40
Alanine aminotransferase (IU/l)	175	7–50
Gamma glutamyl transferase (IU/l)	3	0–8
Total bilirubin (μmol/l)	12.5	0–20
Direct bilirubin (μmol/l)	5.6	0
Indirect bilirubin (μmol/l)	6.9	0–15

showed small pockets of pleural fluid and an area caudal to the heart that was poorly defined and hyperechoic in appearance. The diaphragm could not be clearly identified, ventrally, in any view and thus a diaphragmatic rupture could not be excluded. The cat was anaesthetised on the third day after admission and prepared for a midline coeliotomy and median sternotomy, in the event that the latter were required. During surgery the gall bladder and diaphragm were found to be adhered to each other. This adhesion was dissected to reveal a 2–3 mm defect in the diaphragm (Fig 2) and a corresponding 2–3 mm defect in the gall bladder wall (Fig 2). These defects had formed a sealed fistula between the lumen of the gall bladder and the pleural cavity. There was no evidence of any other abnormalities within the abdomen. The defect in the diaphragm was closed with a cruciate mattress suture of two metric polydioxanone. The defect in the gall bladder was repaired with the above suture material placed in a simple interrupted pattern. The left-sided tube was removed and the coeliotomy incision closed routinely. The pleural cavity was thoroughly lavaged with 0.9% saline through the right-sided thoracostomy tube, and a central venous catheter (Long Term Double Lumen Catheter, Guide Wire Style, 5Fr × 13 cm (5.25 inches), Mila International, Medical Instrumentation for Animals, USA) was placed via the right external jugular vein prior to recovery from anaesthesia for continued fluid therapy, intravenous medications and blood collection when needed.

Minimal effusion was aspirated from the chest drain each day following the surgery, although pleural lavage continued. By the third day the volume of fluid drained from the pleural space, excluding fluid for lavage was 3 ml over a 24 h period. Repeat thoracic radiographs confirmed a significant reduction in the amount of pleural effusion. The right-sided thoracostomy tube was removed on the same day. The tip of the tube was submitted for bacterial culture and again yielded no growth. The cat recovered uneventfully, despite developing a mild upper respiratory tract infection. The cat remained on intravenous amoxicillin–clavulanic acid (Augmentin; GlaxoSmithKline, UK) and metronidazole (Metronidazole; Baxter Health Care, UK) for a further 7 days, when therapy was switched to oral amoxicillin–clavulanic acid (Synulox; Pfizer Animal Health, UK) for a further 14 days. Sutures were removed and the cat was discharged on the 10th postoperative day. Follow-up examinations were conducted 2 and 4 weeks after discharge; on both occasions, no abnormalities were detected and the owner reported the cat to have completely recovered. No abnormalities were detected on repeat abdominal ultrasonography, at the second re-visit.

Biliothorax, also known as bilothorax, is a rare presentation in both dogs and humans and, to the authors' knowledge, has not previously been reported in cats. There are only four cases reported in dogs[1–4]: two of these cases were associated with gunshot injuries and diaphragmatic tears,[1,4] the third was associated with extrahepatic bile duct rupture following a road traffic accident and developed despite an intact diaphragm[2] whilst the most recent case developed postoperatively following cholecystectomy to treat a gall bladder mucocoele, also with an intact diaphragm.[3]

Diagnosis of biliothorax is based on the ratio of the concentration of bilirubin in the pleural effusion to the serum. If the ratio is greater than one then a diagnosis of biliothorax can be made.[5,6] It has been suggested that to support the diagnosis, the appearance of the fluid as well as the bilirubin ratio should be assessed.[3] In this cat, the bilirubin ratio was more than 28 and the appearance of the fluid was bright yellow. Biliothorax

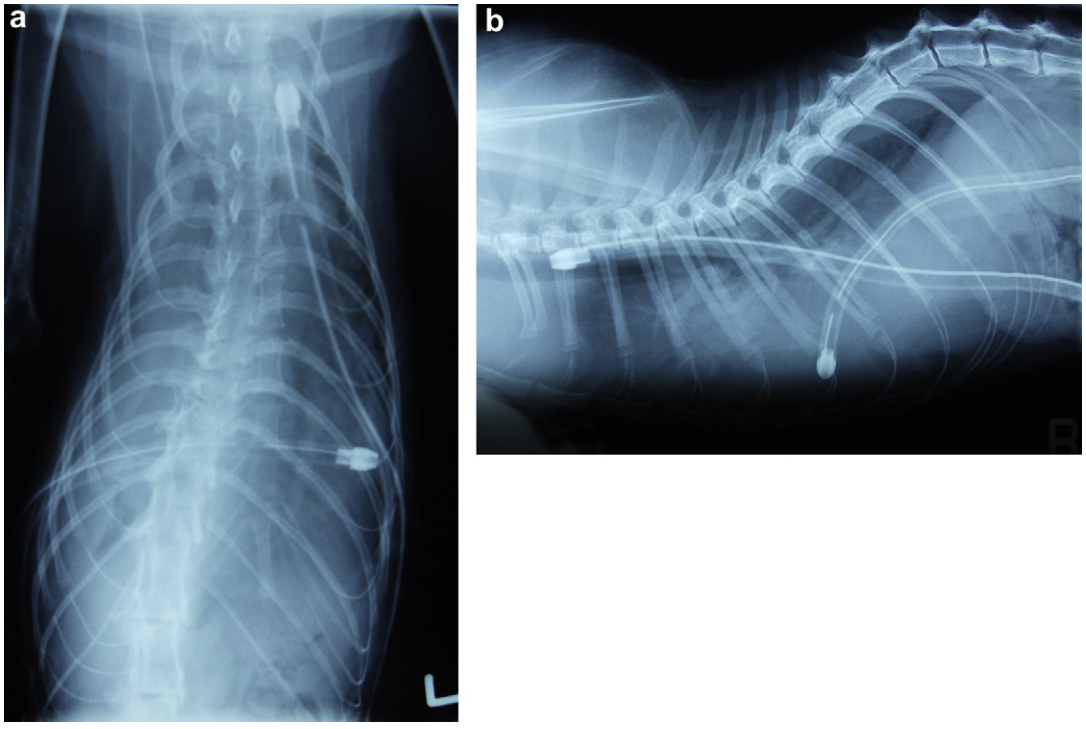

Fig 1. (a) Right lateral thorax radiograph. Note the position of the two thoracostomy drains in the thorax, the moderate pleural effusion and the lack of distinction between the caudal cardiac silhouette and the diaphragm. (b) Dorsoventral thorax radiograph. Note again the position of the thoracostomy tubes, the more severe effusion on the right side and the border effacement of the diaphragm.

can occur in human patients for a variety of reasons and is commonly associated with biliopleural fistulae but can also occur with bile leakage into the abdomen in the face of an intact diaphragm.[5,6] The suspected mechanism of action in the latter scenario is that the bile acids and pigments are carried across the diaphragm in the lymphatics, and then cause damage to the lymphatics, subsequently leaking into the pleural space along with fluid.[1] Fistulae can occur secondary to percutaneous biliary drainage,[5,7,8] liver biopsy,[9] thoracoabdominal wounds, bile duct obstruction, Hodgkin's disease of the liver,[10] hepatic trauma[11] or as a congenital defect.[10] The risk is higher in human patients undergoing various percutaneous procedures where the needle is inserted in the eighth to 10th intercostal space.[12,13] The thoracostomy tube placements in the above case were not ideal. The right tube was inserted in the ninth intercostal space while the left was in the eighth. The procedure has been described in many veterinary texts and it is recommended that the pleura should be penetrated in the seventh–eighth intercostal space.[14–16] Due to the caudal position of the right tube and the absence of any other penetrating trauma or concomitant hepatobiliary pathology, the authors suspect that in attempting to place the tube the referring vet penetrated the caudal pleural cavity then passed the tube through the diaphragm and into the gall bladder before withdrawing and repositioning the tube in the pleural cavity. It is interesting that the gall bladder and diaphragm remained in apposition such that the cat developed a biliothorax rather than a biliary peritonitis. This is not a recognised complication of thoracostomy tube placement but one of the authors (RB) has previously seen biliary aspiration in a cat when attempted drainage of pleural fluid was performed at too caudal a site. Placement at too caudal a location could result in damage to any of the cranial abdominal structures, and the diaphragm.

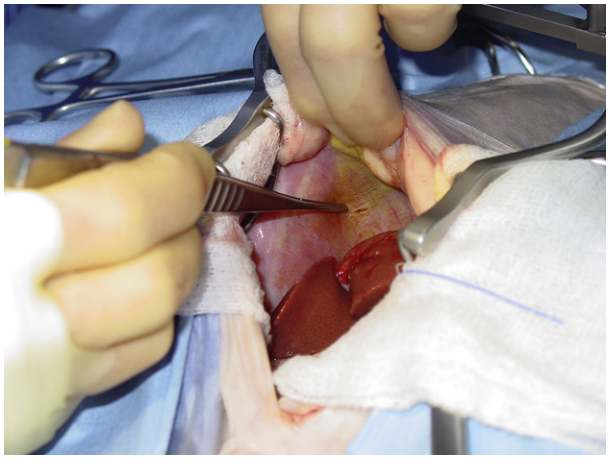

Fig 2. Intra-operative photograph of the defect on the abdominal surface of the diaphragm. The defect corresponded with a defect in the gall bladder. There is bile pigment staining of the diaphragm around the defect.

Extrapolating from the human literature, penetrating trauma to the gall bladder especially where the trauma was directed through the diaphragm could have lead to the biliothorax in the above case. Primary gall bladder pathology with biliothorax has been described in a dog but formed in the face of an intact diaphragm and did not result in fistula.[3] In this case, the gall bladder appeared normal on ultrasonographic examination and during the surgery. However, bile aspiration was not performed at the time, and this may have been helpful in ruling out infectious causes of the fistula formation. It is possible that a fistula formed first between the gall bladder and pleural cavity with a subsequent biliothorax and secondary pyothorax. This has never been reported before in a cat or dog. The four cases previously described, were treated similarly by repair of the defects in the diaphragm, when present, and gall bladder and provision of intermittent pleural space drainage. A significant pleural effusion can be expected in biliothorax due to severe pleuritis caused by contact with bile.[5] This together with a negative acute phase reaction was thought to be the cause of the relative reduced serum albumin concentration on presentation in this case. Although the albumin concentration fell within the reference interval (Table 1), the level of clinical dehydration present on initial examination would suggest that the true concentration may have been lower. Other causes of the reduced albumin concentration such as hepatic dysfunction, protein losing enteropathy or protein losing nephropathy were considered although were not ruled out by laboratory tests. Where biliothorax occurred in the presence of an intact diaphragm the effusion resolved with time.[2,3] Chanoit et al.[3] described the use of prednisolone at a dose of 1 mg/kg q 24 h as part of the treatment in a dog with a biliothorax. The dog had a suspected focal peritonitis following biliary tract surgery in addition to a biliothorax but the reason for use of prednisolone in that patient was unclear from the case report.[3]

The authors' recommendations for managing biliothorax are thoracocentesis, thoracostomy tube placement if necessary, and prompt identification of an underlying cause. Exploratory surgery is indicated if medical management is unsuccessful. In all the published reports of biliothorax in the veterinary literature, as well as this report, the animals were successfully treated with excellent outcomes.

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