Spontaneous external biliary fistula in a dog

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Case Description—A 10-year-old sexually intact female dog was examined because of a static, well-circumscribed subcutaneous mass and associated fistulous draining tract located along the right ventrolateral aspect of the thoracic body wall of 15 months’ duration.

Clinical Findings—Results of computed tomography and fistulography confirmed the presence of the fistulous tract. Computed tomography also revealed a focal, hypodense region in the right ventral portion of the liver that was adjacent to but not clearly associated with the fistulous tract.

Treatment and Outcome—Surgical exploration of the tract revealed that it passed into the right hemithorax to the diaphragm; entered the right medial lobe of the liver; and terminated in a well-encapsulated, cystic liver lesion. The right medial liver lobe and all affected tissues were removed. Histologically, the liver lesion consisted of a fibrotic, dilated bile duct. The dilated bile duct and fistula were lined with biliary epithelium. On the basis of these findings, a diagnosis of spontaneous external biliary fistula was made. Five months after surgery, the dog was clinically normal.

Clinical Relevance—To the authors’ knowledge, spontaneous external biliary fistula in a dog has not been reported in the veterinary medical literature. Despite the rarity of this condition, it should be considered in a dog with similar clinical findings. Clinical findings and results of appropriate diagnostic imaging procedures may provide valuable information in making this diagnosis and in planning surgical treatment. (J Am Vet Med Assoc 2007;231:919–923)

A 10-year-old 20.4-kg (44.9-lb) sexually intact female Bearded Collie was referred to the UFVMC with a 15-month history of a well-circumscribed, 3 × 5 × 1-cm, subcutaneous mass and associated purulent, fistulous draining tract located along the caudal right ventrolateral portion of the thoracic body wall. The dog had not had signs of illness and had apparently normal appetite and activity level.

Twelve months prior to referral, a similar but non-draining mass was evaluated by the referring veterinarian. The mass was removed, and the dog was discharged by the referring veterinarian and treated with a 4-week course of cephalexin (25 mg/kg [11.4 mg/lb], PO, q 8 h). Six weeks after the mass was removed, the dog was returned to the referring veterinarian because of recurrence of the mass; drainage from the mass had begun at that time. A 5-mm punch biopsy specimen was obtained, and histologic examination revealed mild to moderate chronic lymphoplasmacytic cellulitis, accompanied by fibrosis. Loose collagen fibers with blood vessels and occasional skeletal muscle cells were mixed with perivascular lymphocytes, plasma cells, and edema. No bacteria or fungi were identified with special stains. Bacteriologic culture resulted in scant growth of oxacillin-resistant *Staphylococcus intermedius* that was susceptible to enrofloxacin. There was no evidence of mycobacterial or fungal growth. Results of a serum reference limits. Because of the highly resistant bacteria...

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causing the infection, antimicrobial treatment was discontinued and surgical exploration and resection were performed.

Preoperative fistulography and CT of the caudal portions of the thorax and abdomen were performed. Computed tomographic images were reconstructed into 3-mm-thick slices. A 6-F Foley catheter was placed in the external portion of the draining tract along the body wall. The Foley bulb was filled with 3 mL of air. Iohexol (1.2 g) was injected into the draining tract, and the imaging procedure was repeated. Focal thickening of the subcutaneous tissues was observed along the right ventral portion of the thoracic and abdominal body wall beginning at the level of the 6th rib and extended approximately 10 cm caudally to the level of the 10th costal cartilage. The thickened tissue increased in size caudally and measured approximately 2 cm in depth at its thickest point. The lesion was approximately 5 cm, as measured dorsoventrally. Fistulography revealed a sinus tract extending caudally and dorsally and containing multiple irregularly marginated saccular dilations (Figure 1). There was no conclusive evidence of a foreign body, but a focal hypodense region with a slightly mineralized center was detected in the right ventral portion of the liver (Figure 2). Contrast medium was identified medial to the ribs close to this region, but it could not be definitively determined whether the hypodense region communicated with the cutaneous fistulous tract.

Prior to surgery, methylene blue (diluted 1:3 with sterile water) was injected into the fistulous tract to facilitate identification of its margins during surgery. An elliptical incision, encircling and incorporating all of the affected tissue of the thoracic body wall, was made to the level of the ribs. Substantial hemorrhage was immediately encountered from indistinct vessels coursing through severely inflamed, highly vascular granulation tissue in the subcutis. Following en bloc resection of

![Figure 1](https://example.com/image1.png)

**Figure 1**—Ventrudorsal (A) and lateral (B) radiographic views (obtained after administration of contrast agent) of the thoracic and abdominal regions of a dog with a spontaneous external biliary fistula that extends caudally and dorsally and has multiple irregularly marginated saccular dilations. The fistula appears to terminate at the distal aspect of the right 10th rib at the level of the costochondral junction.

![Figure 2](https://example.com/image2.png)

**Figure 2**—Computed tomographic image of the abdomen of the dog in Figure 1 at the level of the 10th rib. Notice the fistula and the hypodense cystic lesion in the right ventral portion of the liver (arrowhead).
the affected skin and subcutis, a small tract traversing through the body wall was evident because of methylene blue at the 10th intercostal space. A paracostal thoracotomy was made at the intercostal space between the 10th and 11th ribs. The tract was associated with a large adhesion between the diaphragm and right medial liver lobe. The right medial liver lobe was isolated, and a partial liver lobectomy was performed with a 90-mm thoraco-abdominal stapler that used a double row of 3.5-mm staples. The affected portion of diaphragm and associated adhesion were removed with the liver lobe. The remainder of the liver and gallbladder appeared grossly normal. The cystic and common bile ducts were patent. Once exteriorized, the liver lobe was more closely explored. Within the resected liver tissue, a fibrous, 1.5-cm-diameter, cystic structure that contained moist, chalky, friable, greenish-black material was detected (Figure 3).

Prior to closure, hemorrhage from the remainder of the right medial liver lobe or from elsewhere in the thorax or abdomen was not observed. The internal thoracic artery was intact. The diaphragmatic defect was closed in a simple continuous pattern, and the diaphragmatic margin was sutured to the 11th rib. A thoracostomy tube was placed routinely prior to closure of the thoracic cavity. The remainder of the surgical site was closed routinely. Bleeding from the subcutaneous tissues persisted during closure.

Initial postoperative management included administration of hydromorphone (0.01 mg/kg/h [0.005 mg/lb/h], IV via continuous rate infusion), amikacin (20 mg/kg, IV, q 24 h), metronidazole (15 mg/kg [6.8 mg/lb], IV, q 24 h), and carprofen (2 mg/kg [0.91 mg/lb], SC, q 24 h); central venous pressure measurements every 4 hours; and evacuation of the thoracostomy tube with measurement of fluid volume every hour. After surgery, the subcutis continued to bleed through the skin incision as well as into the thorax. The blood lost into the thoracic cavity was evacuated via the thoracic tube. The hemorrhage resolved within 24 hours with the use of a pressure bandage and ice packing. A whole blood transfusion was also provided after surgery to maintain the Hct, which had decreased from 46% before surgery to 33% immediately after surgery and 20% at 8 hours after surgery.

Bacteriologic culture and susceptibility testing of the contents of the cystic liver lesion revealed moderate growth of 4 strains of fluoroquinolone-resistant E. coli. The oxacillin-resistant beta-hemolytic Staphylococcus spp cultured from the cutaneous fistula prior to surgery were not cultured from the liver lesion. On the basis of the culture and susceptibility results, the dog was discharged 3 days after surgery and was treated with amoxicillin-clavulanic acid (20 mg/kg, PO, q 12 h for 2 weeks). Five-month follow-up examination revealed that the mass had not returned, the drainage had resolved, and the dog was clinically normal.

Histologic examination of the resected portion of liver revealed a locally extensive region of disrupted hepatic architecture characterized by loss and replacement of hepatocytes with loosely arranged fibrous connective tissue and marked biliary and vascular proliferation. The cystic liver lesion was partially lined by columnar to cuboidal epithelium with goblet cells and resembled biliary epithelium. Surrounding the epithelium were bundles of organized smooth muscle that merged with granulation tissue. Extensive areas of erosion and ulceration of the epithelium were evident along with a bright golden granular material presumed to be bile. Nodular infiltrates of lymphocytes and fewer plasma cells were within the lamina propria of the surrounding connective tissue. The origin of the fistula appeared to be a large, dilated, fibrotic bile duct that contained inspissated bile mixed with cellular debris and hemorrhage. Hepatic degeneration, localized chronic ectatic cholangitis, and biliary and vascular hyperplasia were also present. Adjacent liver tissue was markedly degenerate, fibrotic, and hemorrhagic, which was likely associated with the local inflammation and bile retention. No foreign body was identified.

The cutaneous draining tract was lined with epithelium resembling that of the bile duct. The tract extended from the epidermis into and through the panniculus adiposus, which was replaced by granulation tissue with numerous, scattered, small lymphoid aggregates. Immature vascular granulation tissue containing dense infiltrates of large macrophages with foamy eosinophilic cytoplasm, lymphocytes, plasma cells, and neutrophils was mixed with abundant eosinophilic cellular debris and fibrin. The tract also contained fragments of disorganized, hyperplastic epithelium with substantial variability of the individual cells. Small fluid aggregations (presumed to be bile) were surrounded by neutrophils. No infectious organisms were identified.

Histopathologic findings and clinical observations were nearly identical to a condition in humans called spontaneous external biliary fistula. Consequently, a final diagnosis of spontaneous external biliary fistula was made.

Discussion

To the authors’ knowledge, this is the first report of spontaneous external biliary fistula formation in an adult dog. In humans, external biliary fistulae or cholecystocutaneous fistulae have been described as most commonly occurring in 50- to 70-year-old women, who

Figure 3—Photograph of a portion of the right medial liver lobe of the dog in Figure 1. Notice the fibrous, dilated bile duct that was the origin of a spontaneous external biliary fistula. The contents of the bile duct have been removed. Bar = 1.5 cm.
have greater prevalence of cholecystitis, compared with men. \(^1\) A similar sex predilection with regards to cholecystitis has not been identified in dogs.

As evident in the dog in this report, the disease in humans typically involves a single, painless, enlarging mass that ruptures to reveal a draining sinus tract in the right upper quadrant of the abdomen. \(^1\) Although most external biliary fistulae exit in the right upper quadrant, they may also exit in the flank, thigh, gluteal region, or umbilicus. \(^1\) Cholecystic fistulae may also form internally and involve viscera, with no external communication. \(^3\)

External biliary fistulae in humans have almost always been reported as secondary to neglected biliary tract disease, abdominal trauma, previous abdominal surgery, cholelithiasis, malignancies of the gallbladder, or cholecystitis. \(^1\) Interestingly, external biliary fistulae are typically not noted by an obvious episode of acute cholecystitis, and fistula formation may go unnoticed until the development of a cutaneous exit. \(^3\) The abdominal wall lesion that ultimately forms may be confused with a pyogenic granuloma, infected epidermal inclusion cyst, retroperitoneal tumor, or metastatic carcinoma, frequently leading to misdiagnosis. \(^7\) Presently, spontaneous external biliary fistula formation is considered a clinical rarity in humans because of improvements in modern diagnostic techniques, superior biliary surgical techniques, and an increase in the number of elective cholecystectomies performed. \(^1\) \(^6\) \(^13\)

Computed tomography and fistulography may play important roles in determining the presence and course of the fistula and in confirming the communication of a cutaneous lesion with the liver, gallbladder, or biliary tract. Both can be invaluable in formulation of treatment recommendations, including surgical planning for any chronically draining or recurring tracts. \(^3\) \(^13\) \(^14\) \(^16\)

It is important to note that in the case reported here, CT images had been reconstructed into 3-mm-thick slices. Although CT imaging was performed before and after administration of a contrast agent, it could not be definitively determined whether the hypodense region in the liver communicated with the cutaneous fistulous tract. Smaller CT slices may have provided more accurate information.

Umbilico-biliary fistula formation in a dog, from a preexisting congenital malformation between the biliary system and the umbilicus, has been reported. \(^1\) \(^4\) That congenital fistula coursed cranially from the umbilicus and emptied into the hepatic duct of the right lateral liver lobe at the level of the common bile duct. Similar to the dog of this report, biliary epithelium lined the fistula and the epithelial-lined tissue formed a large duct-like structure that drained bile. \(^1\) \(^4\)

Cause of the spontaneous external biliary fistula in the dog of this report was unknown. In the liver, bile drains through the canaliculi into interlobular ducts that unite to form hepatic ducts; the hepatic ducts emerge from the lobes of the liver and empty into the cystic duct, which ultimately drains into the gallbladder. \(^1\) \(^7\) If there were no link to the main biliary tree or failure of external drainage of accumulated bile, a biliary cutaneous fistula could develop. \(^6\)

Biliary stasis has been suggested as a predisposing factor for formation of biliary sludge in the gallbladder; biliary sludge may be a factor in formation of gallbladder mucocoeles in dogs. \(^9\) In humans, after liver transplantation, mucocoele formation has occurred because of cystic duct remnants that form blind pockets in the biliary tree. \(^20\) Hepatic resection in humans has also been reported to cause intractable biliary leakage from an isolated bile duct in the remaining portion of the liver, resulting in a biloma with biliary-cutaneous fistula formation. \(^9\) In the dog of this report, the cystic structure identified in the right medial liver lobe contained inspissated bile. Although the chalky texture of the material within the liver lesion was unlike that of a classical mucocoele, it is conceivable that biliary stasis that caused biliary sludge, or an isolated duct without communication with the main biliary tree, resulted in formation of an atypical biliary mucocoele or biloma.

In humans, external biliary fistula formation is also associated with chronic gallbladder rupture caused by ongoing, often asymptomatic calculous or acalculous cholecystitis. \(^1\) \(^3\) \(^6\) \(^8\) \(^9\) \(^10\) Inflamed tissue that results from the rupture becomes walled off; a localized cholecystic abscess forms; and, over time, a fistulous tract develops. \(^13\) In the dog reported here, however, the gallbladder had no gross lesions, was fully patent, and had no apparent association with the cystic structure or fistulous tract. The dog also had no serum biochemical abnormalities that would suggest cholestasis. A fecal sample obtained from the dog before surgery yielded negative results for fluke ova via sedimentation, and the dog had no known history of severe trauma; however, one may speculate that an episode of severe focal inflammation caused by a subclinical liver fluke infestation or unnoticed acute abdominal trauma may have occurred.

The persistent hemorrhage in this case may have been unique to the dog or may be an important component of this condition. Histologic examination revealed extensive vascularization of the subcutis, which was consistent with the severe hemorrhage encountered during surgery. Such vascularity could be attributed to the chronic inflammation that is often associated with bile leakage into surrounding soft tissues. \(^3\) \(^13\) \(^16\) Because of the continuous bleeding, all major vascular structures in the surgical site were carefully inspected and were intact. The only source of bleeding that could be identified was the subcutis, and electrocautery and prolonged application of pressure were largely ineffective. Use of surgical fibrin sealant was considered and may have been effective in controlling the hemorrhage but was not available at the time of surgery. After surgery, with ice-packing, application of a pressure bandage, and sufficient time, the bleeding stopped and the dog made a complete recovery. In humans, acute inflammation of the affected tissue is often controlled prior to surgery by use of appropriate antimicrobial treatment after bacteriologic culture of inflamed tissues. After resolution of the acute inflammation, surgical excision is performed. \(^3\) \(^13\) \(^16\) In the dog of this report, a diagnosis had not been made and, consequently, antimicrobial treatment was not considered prior to surgery. In retrospect, however, administration of an appropriate antimicrobial before surgery may have been beneficial.

Although this condition in an adult dog has apparently not been reported, the diagnosis and surgical cor-
rection in this case were reasonably straightforward. On the basis of this case, the prognosis appears to be excellent for complete resolution. Despite its rarity, spontaneous external biliary fistula should be considered in a dog that has a discharging sinus in the right cranial portion of the thoracic or abdominal wall and an associated liver lesion. Clinical findings, analysis of draining fluid, and fistulography with CT or magnetic resonance imaging may provide valuable information in making the diagnosis and planning surgical treatment.

References