Surgical Management of Biliary Duct Hamartoma in a Cat: A Case Report

Nur Ainina Ab Manap[1], Tey Yu Chong[2], Erni Wati Mohd Arip[2], Rozanaliza Radzi[2], Muhamad Alif Zakaria[3], Nur Diyana Mohamad Tahir[4], Nurul Izzati Uda Zahli[5]*

[1]Faculty of Veterinary Medicine, Universiti Putra Malaysia, 43400 UPM Serdang, Selangor, Malaysia, [2]Department of Companion Animal Medicine and Surgery, Faculty of Veterinary Medicine, Universiti Putra Malaysia, 43400 UPM Serdang, Selangor, Malaysia, [3]Department of Veterinary Laboratory Diagnostic, Faculty of Veterinary Medicine, Universiti Putra Malaysia, 43400 UPM Serdang, Selangor, Malaysia, [4]Department of Veterinary Clinical Studies, Faculty of Veterinary Medicine, Universiti Putra Malaysia, 43400 UPM Serdang, Selangor, Malaysia, [5]Department of Veterinary Pathology and Microbiology, Faculty of Veterinary Medicine, Universiti Putra Malaysia, 43400 UPM Serdang, Selangor, Malaysia.
*Corresponding author: nurulizzati.udazahli@upm.edu.my

Abstract

Cysts in the liver or biliary duct are uncommon in veterinary medicine. A multiloculated, fluid-filled liver cyst measuring 18 cm in diameter was detected in a two-year-old spayed female mixed-breed cat via radiography and computed tomography. The cyst was attached to the medial lobe aspect of the liver and continued with the gall bladder. Cystectomy and omentopexy were performed, and the resected cyst was examined histologically. Histologic analysis revealed variable-sized cystic spaces lined by low simple cuboidal and attenuated epithelium. The cyst wall was composed of thick collagenous stroma containing entrapped islands of the hepatic parenchyma, which included atrophied hepatocytes, dilated sinusoidal spaces filled with erythrocytes, and randomly distributed hyperplastic bile ducts. These histologic findings were consistent with biliary duct hamartoma. The cat had an uneventful recovery, and no recurrence was observed one-year post-surgery.

Keywords: bile duct hamartoma, cystectomy, ductal plate malformation, hepatic cyst, von Meyenburg complex

Received: October 16, 2024 **Revised:** January 2, 2025 **Accepted:** February 3, 2025

INTRODUCTION

Biliary duct hamartoma (BDH), also known as von Meyenburg complex, bile duct hamartoma, or biliary hamartoma, is a congenital cystic lesion resulting from ductal plate malformation (DPM). A ductal plate is defined as a double-layered cylindrical structure of bile duct epithelium that surrounds the portal vein ramifications during gestation, which will develop and remodel into intrahepatic bile ducts (Venkatanarasimha *et al.*, 2011). Disruption in the developmental and remodeling phases of the ductal plate leads to a continuum of lesions grouped as ductal plate malformation.

In human medicine, fibropolycystic disease is an umbrella term used for a spectrum of conditions resulting from DPM, and this categorization is considered applicable to veterinary medicine (Center, 2023). Across animal species, BDH is uncommon and reported sporadically in mammals and reptiles (Naghi *et al.*, 2023). Given the rarity of hepatic cysts, this case description is valuable for veterinarians to consider BDH as a differential diagnosis when encountering patients with hepatic cysts.

Surgical management of BDH is important, mainly when cysts grow large enough to compress surrounding structures or cause clinical signs. Options for surgical intervention include partial or total liver lobectomy and hepatic cystectomy, depending on the cyst's size, location, and adherence to adjacent structures (Radlinsky and Fossum, 2019). The surgical techniques employed in this case contribute to understanding BDH management in veterinary practice.

RESULTS AND DISCUSSION

Case Description

A two-year-old spayed female mixed-breed cat was referred to the university veterinary hospital for cystectomy of a liver cyst. The abdominal cyst had been discovered incidentally during an ovariohysterectomy one year earlier. A year later, radiography and computed tomography conducted at a private animal hospital identified a large, multiloculated, fluid-filled cyst approximately 18 cm in diameter, attached to the liver (Figures 1 and 2). No abnormalities were noted in the rest of the liver, kidneys, or other organs.

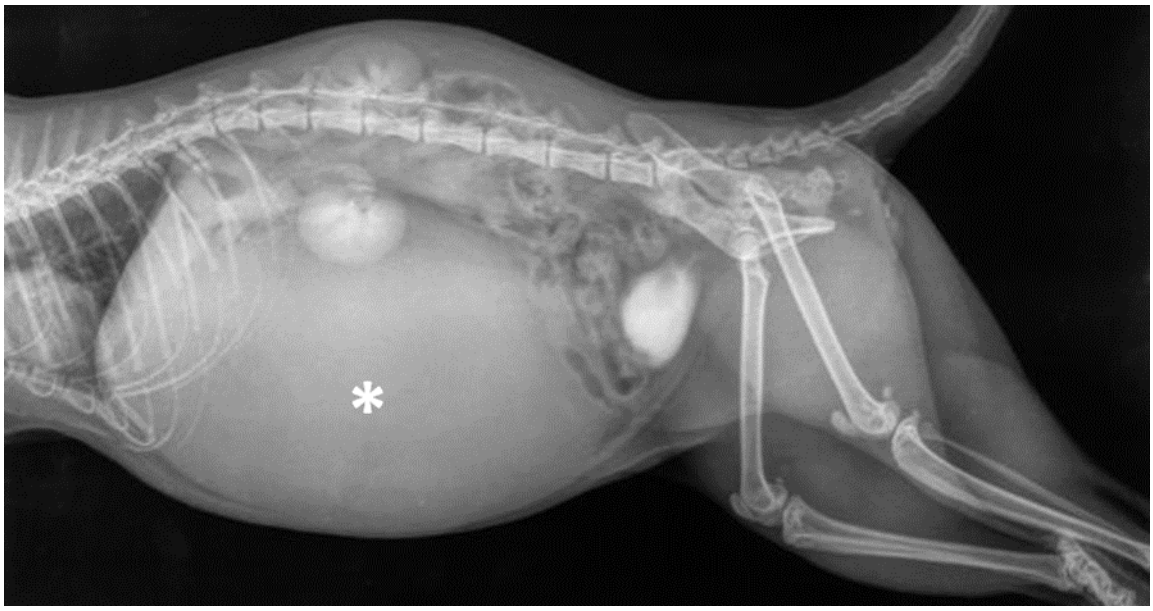

Figure 1. Radiographic image showing a significant, well-demarcated soft tissue or fluid opacity in the abdominal cavity, causing craniodorsal displacement of the intestines (asterisk).

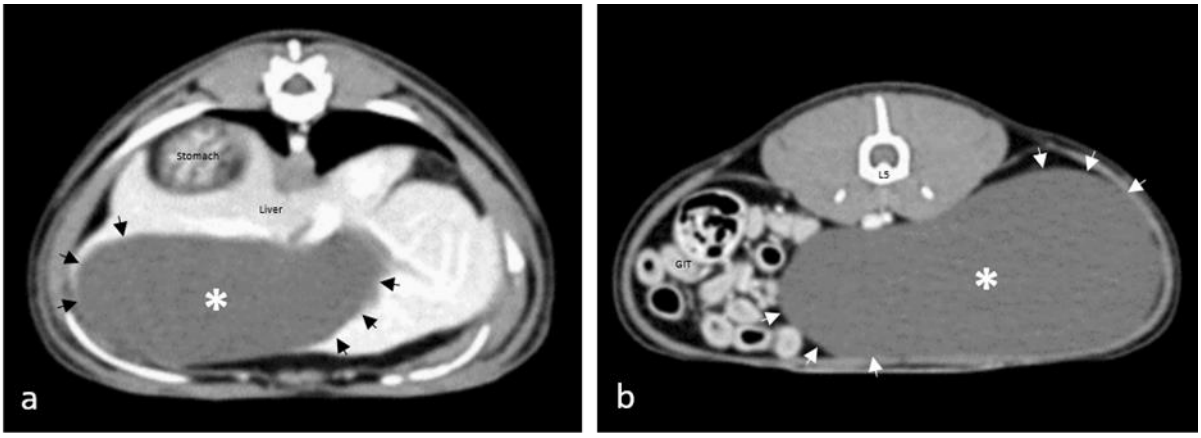

Figure 2. Computed tomography images showing a homogeneous, hypoattenuating, encapsulated (arrows) area of fluid to soft tissue density (asterisk), with (a) liver attachment and (b) intestinal tracts displaced to the left side of the abdominal cavity at L5 level.

Upon physical examination, the cat was quiet, alert, and responsive, with no abnormalities detected in basic parameters. The cat showed symmetrical abdominal enlargement and abdominal pain when the abdomen was palpated. Hematological analysis revealed a stress leukogram with mild neutropenia (2.22×10^9/L; reference range $2.30–10.29 \times 10^9$/L) and mild hyperglycemia (10.57 mmol/L; reference range 4.11–8.84 mmol/L). The differential diagnosis

included benign growths such as hepatic cysts and biliary cystadenoma, prompting exploratory laparotomy for hepatic cystectomy.

The cat received intravenous premedication with tramadol (2 mg/kg) and midazolam (0.2 mg/kg). Anesthesia was induced with propofol (5 mg/kg) intravenously and maintained with isoflurane at 1.5–2%. The cat was positioned in dorsal recumbency, and a skin incision was made from the sternum to the umbilicus, followed by incisions through the subcutaneous tissue, linea alba, and peritoneum. The cyst had spontaneously ruptured, releasing serosanguineous fluid into the peritoneal cavity, which was aspirated using suction, with a total volume of 38.4 mL. One intact cyst was observed, and its fluid was aspirated using a 10 mL syringe and a 25G needle. The attachment between the cyst and the liver was then assessed. The cyst wall had extensive attachments to the visceral surfaces of the left medial lobe, left lateral lobe, right medial lobe, and gallbladder (Figure 3).

The cyst's attachment to the liver and gallbladder was carefully dissected, and bleeding was controlled using bipolar and unipolar cauterizers. Omentopexy was then performed by suturing the greater omentum to the remaining cyst wall still attached to the liver, using PDS 4/0 in a simple interrupted suture pattern. Before closing the incision, the abdomen was lavaged with warm normal saline (1 L, 250 mL/kg), and the fluid was removed by suction. The linea alba and peritoneum were closed with a simple continuous suture pattern, while the subcutaneous layer was closed with a modified Cushing pattern using PDS (3/0). The skin was closed with an intradermal suture pattern using Vicryl (3/0). Fluid from the intact cyst was sent for microscopic examination and bacterial culture, which revealed no significant findings. The resected cyst capsule was sent for histopathological examination.

For postoperative pain management, the cat received meloxicam (0.1 mg/kg subcutaneously) once daily for three consecutive days. The cat was discharged eight days after surgery without complications. Follow-ups at 11 days, 26 days, 295 days, and one year post-surgery revealed no cyst recurrence on abdominal radiographs.

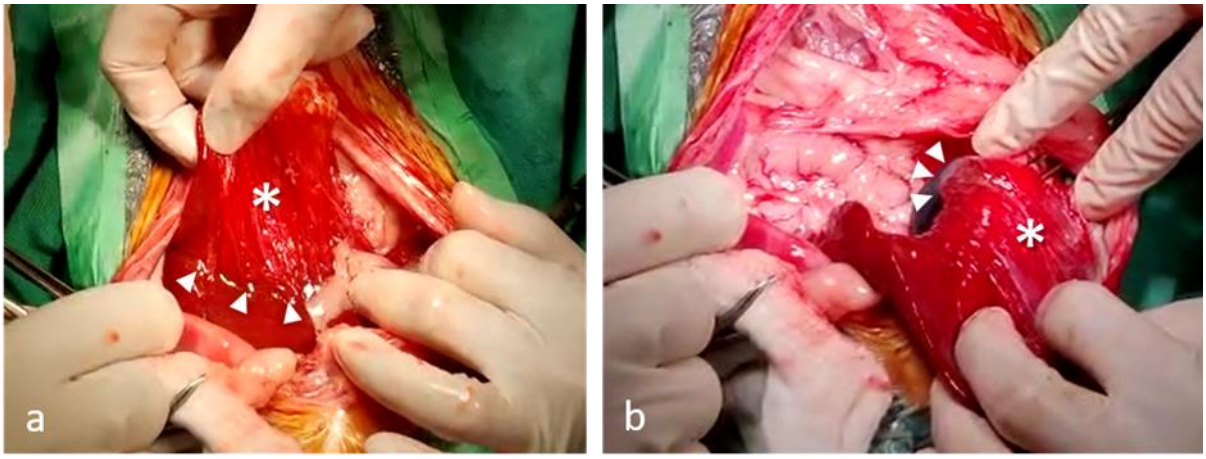

Figure 3. The cyst wall (asterisk) is continuous with (a) the medial lobe of the liver (arrowheads) and (b) the gallbladder wall (arrowheads). The cyst has spontaneously ruptured, releasing 38.4 mL of clear serosanguines fluid.

The extracted cyst was fixed in 10% buffered formalin and submitted for routine histologic processing. Histologically, the specimen consisted of several large, extensively dilated, and small cystic spaces lined by low simple cuboidal and attenuated epithelium. The cyst wall was composed of thick collagenous stroma containing scattered foci of lymphoplasmacytic infiltration (Figure 4a). The lumens contained amorphous, lightly eosinophilic material in the smaller cystic spaces. Randomly distributed throughout the cyst wall were islands of entrapped hepatic

parenchyma. The hepatocytes were often shrunken (atrophied), and the sinusoidal spaces were dilated and filled with erythrocytes (Figures 4b and 4c). Portal triads were present but moderately disrupted. Hyperplastic bile ducts were also randomly distributed within the cyst wall (Figure 4d). These findings are consistent with a diagnosis of BDH.

Discussion

It is crucial to differentiate liver and biliary cysts from acquired cysts and developmental or congenital disorders for accurate treatment. Acquired cysts, such as those seen in *Platynosomum* spp. and *Echinococcus* spp. infections must be distinguished from congenital cystic disorders, including simple hepatic or biliary cysts, BDH, Caroli malformation, choledochal cysts, and biliary cystadenomas (Andrade *et al.*, 2012; Bonelli *et al.*, 2018; Center, 2023). There was no evidence of helminths or associated inflammatory reactions, ruling out acquired causes in this case. Congenital cysts, including simple hepatic or biliary cysts, choledochal cysts, and biliary cystadenomas, typically lack hepatic parenchyma embedded in the cyst wall (Kim *et al.*, 2021; Schreeg *et al.*, 2021; Moon *et al.*, 2011).

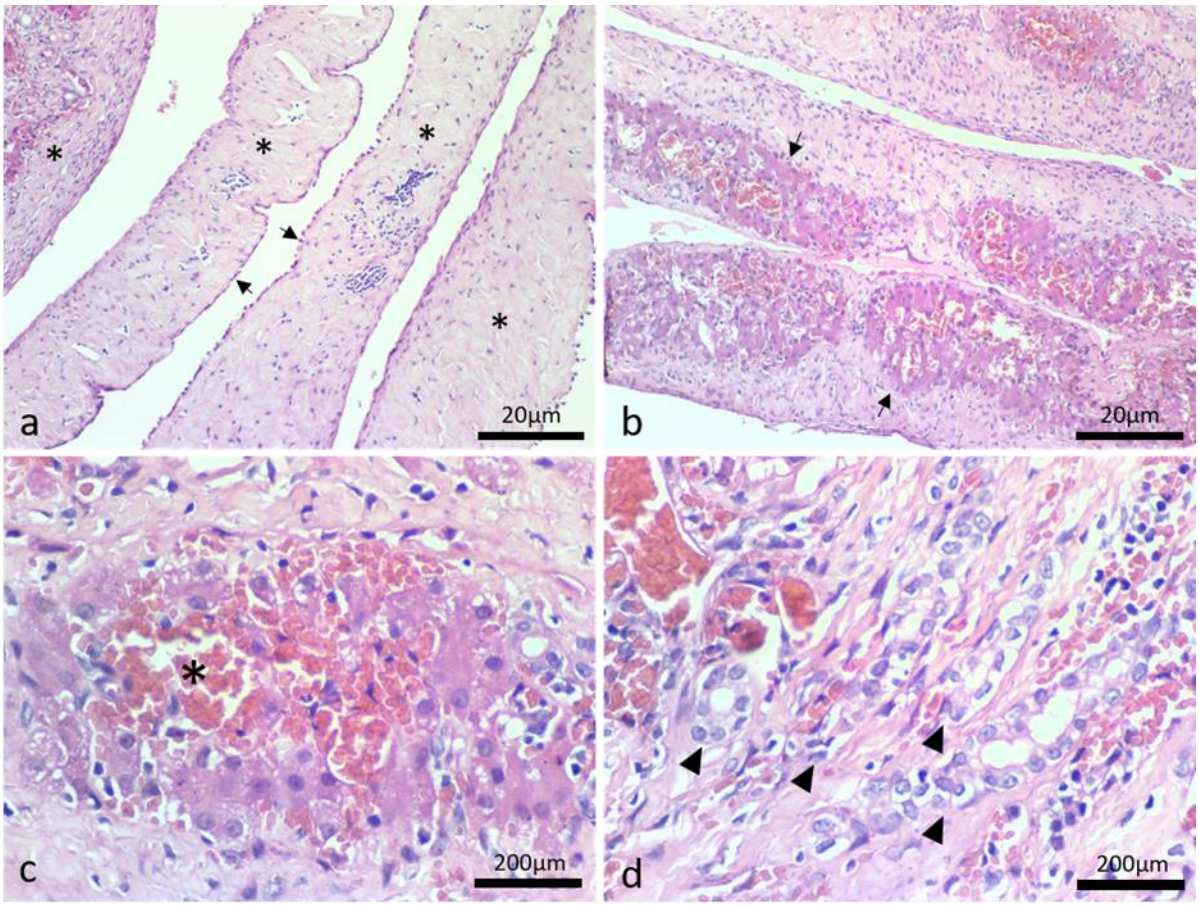

Figure 4. (a) The cyst wall is composed of thick collagenous stroma (asterisks) with scattered foci of lymphoplasmacytic infiltration, lined by low simple cuboidal and attenuated epithelium (arrows). HE, 80×. (b) Islands of hepatic parenchyma (arrows) entrapped within the cyst wall. HE, 80×. (c) Shrunken hepatocytes with dilated sinusoidal spaces filled with erythrocytes (asterisk). HE, 800×. (d) Hyperplastic bile ducts (arrowheads) are randomly distributed within the cyst wall. HE, 800×.

The solitary space-occupying lesion, in this case, does not alter the hepatic parenchyma or cause intrahepatic bile duct dilatation, in contrast to findings in Caroli-type malformation (Roberts *et al.,* 2018). The diagnosis of BDH in this case was challenging due to the cyst's considerable size and the absence of the typical "honeycomb" histologic appearance. However, a key histopathological feature of BDH is the entrapment of hepatic parenchyma within the cyst wall.

The possible causes of BDH, as with other DPMs, involve disruptions during the developmental and remodeling phases of the ductal plate in utero (Venkatanarasimha *et al.*, 2011). These disruptions can result from genetic or environmental factors, such as maternal malnutrition, toxins, or infections during gestation, although the exact etiology in this case remains unclear. BDH is typically small in humans, with an average size of 1 cm based on 139 cases (Sheikh *et al.*, 2022). In animals, the size of BDH cysts varies from 7 cm in a cat (Naghi *et al.,* 2023), 8.7 cm in a dog (Mao *et al.*, 2023), 7 cm in a calf (De Bosschere *et al.*, 1999), and 4 cm in a rabbit (Starost *et al.*, 2007). Despite its size, BDH is generally asymptomatic and is often detected only during postmortem examinations or incidentally during imaging procedures, such as ultrasound or CT scans, as in this case. The initial cyst size, in this case, was unknown, but over time, the cyst either enlarged or coalesced to form a sizeable space-occupying mass.

While BDH is typically benign and does not impair liver function, large cysts can compress surrounding tissues, leading to clinical signs such as vomiting or abdominal distention. Complications such as ascites or adhesions to peritoneal organs might follow if ruptured. Rarely have there been cases where BDH progressed to malignancies such as intrahepatic cholangiocarcinoma or hepatocellular carcinoma (Sheikh *et al.,* 2022). As a result, surgical excision is typically recommended for both therapeutic and preventive purposes.

Partial hepatectomy is the most prevalent treatment method for BDH (Sheikh *et al.*, 2022). Total or partial lobectomy is not indicated in this case due to its benign nature and extensive adhesion to the liver capsule and gall bladder. Instead, hepatic cystectomy was performed as a less invasive alternative, accompanied by omentopexy, which enhances vascularization by providing angiogenic and phagocytic properties and minimizes postoperative complications such as bleeding or fluid accumulation (Naga *et al.,* 2020). The recovery was excellent in this case as the cat demonstrated uneventful healing without recurrence or postoperative complications a year post-surgery. The absence of recurrence on abdominal radiographs and the resolution of clinical signs, such as abdominal pain and distension, indicate that the surgical approach was practical.

Although the short-term and medium-term outcomes were favorable, long-term follow-up beyond one year is unavailable. Thus, the potential for late recurrence or complications is unknown. Given the rarity of BDH, the findings from this single case may not be generalizable to all cats with similar presentations. Future studies involving case series are necessary to support the efficacy of the described surgical techniques in managing BDH in veterinary medicine.

CONCLUSION

Although rare, the diagnosis of BDH should be included as one of the differential diagnoses for liver cysts in young cats, and it can be successfully managed with cystectomy and omentalization in veterinary practice.

ACKNOWLEDGEMENTS

The authors would like to express their gratitude to the University Veterinary Hospital and the Histopathology Laboratory at the Veterinary Laboratory Service Unit, UPM, for providing the case and for their excellent service.

AUTHORS' CONTRIBUTIONS

NAAM: Conceptualization and writing original draft; TYC, EWMA, RR and NDMT:

Supervising student, perform analysis and case investigation; NIUZ: Review, writing and editing final draft. All authors have read, reviewed, and approved the final manuscript.

COMPETING INTERESTS

The authors declare that they have no competing interests.

REFERENCES

Andrade, R. L., Dantas, A. F., Pimentel, L. A., Galiza, G. J., Carvalho, F. K., Costa, V. M., & Riet-Correa, F. (2012). Platynosomum fastosum-induced cholangiocarcinomas in cats. *Veterinary Parasitology*, 190(1–2), 277–280.

Bonelli, P., Masu, G., Dei Giudici, S., Pintus, D., Peruzzu, A., Piseddu, T., Santucciu, C., Cossu, A., Demurtas, N., & Masala, G. (2018). Cystic echinococcosis in a domestic cat (*Felis catus*) in Italy. Échinococcose kystique chez un chat domestique (Felis catus) en Italie. *Parasite (Paris, France)*, 25, 25.

Center, S. A. (2023). 'Miscellaneous Disorders of the Bile Ducts in Small Animals', in *MSD Veterinary Manual*. Available: https://www.msdvetmanual.com. (Accessed: 30th June 2024)

De Bosschere, H., & Ducatelle, R. (1999). Bile duct hamartoma in a calf. *The Veterinary Record*, 144(8), 210–211.

Kim, K., Kim, H., Eom, K., & Kim, H. (2021). Surgical management and long-term follow-up of a giant hepatic cyst with an internal septum in a cat. *Journal of Veterinary Clinics*, 38(6), 295–298.

Mao, D., Song, X., Ma, D., Hu, S., Zhang, Z., Wang, J., & He, X. (2023). Bile duct hamartoma in a dog. *Journal of Comparative Pathology*, 207, 45–49.

Moon, S. J., Kim, J. W., Sur, J. H., Jeong, S. W., & Park, H. M. (2011). Biliary cystadenoma in a Maltese dog: clinical and diagnostic findings. *The Journal of Veterinary Medical Science*, 73(12), 1677–1679.

Naga, M. A., Abd El Aal, A. A., Mousa, A. S., & Saber, H. S., (2020). Omentopexy in sleeve gastrectomy and its effect on postoperative complications. *The Egyptian Journal of Surgery*, 39(4), 1208.

Naghi, R., Bertran, J., Spoldi, E., Dark, M. J., de Oliveira, H. H., Souza, C., & Maxwell, E. A. (2023). Multiple biliary duct hamartomas in a cat resulting in a hepatic mass: A case report. *Veterinary Medicine and Science*, 9(4), 1441–1445.

Radlinsky, M. A., & Fossum, T. W. (2019). Surgery of the liver. In Fossum, T. W. (Ed) Small Animal Surgery. 5th edition. Philadelphia PA: Elsevier. pp: 540–570.

Roberts, M. L., Rine, S., & Lam, A. (2018). Caroli's-type ductal plate malformation and a portosystemic shunt in a 4-month-old kitten. *JFMS Open Reports*, 4(2), 2055116918812329.

Schreeg, M. E., Miller, S. A., & Cullen, J. M. (2021). Choledochal cyst with secondary cholangitis, choledochitis, duodenal papillitis, and pancreatitis in a young domestic shorthair cat. *Journal of Veterinary Diagnostic Investigation*, 33(4), 782–787.

Sheikh, A. A. E., Nguyen, A. P., Leyba, K., Javed, N., Shah, S., Deradke, A., Cormier, C., Shekhar, R., & Sheikh, A. B. (2022). Biliary Duct Hamartomas: A Systematic Review. *Cureus*, 14(5), e25361.

Starost M. F. (2007). Solitary biliary hamartoma with cholelithiasis in a domestic rabbit (*Oryctolagus cuniculus*). *Veterinary Pathology*, 44(1), 92–95.

Venkatanarasimha, N., Thomas, R., Armstrong, E. M., Shirley, J. F., Fox, B. M., & Jackson, S. A. (2011). Imaging features of ductal plate malformations in adults. *Clinical Radiology*, 66(11), 1086–1093.

***