

## Case Report

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# Von Meyenburg Complex in a Cat and a Dog, and Mini Literature Review

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### ABSTRACT

Biopsies of liver from a 10-year-one-month-old male, neutered dog and an adult, male, domestic short haired cat of unknown age were received and microscopically examined. In both cases ectatic bile ducts consistent with von Meyenburg complex also known as bile duct hamartoma (BDH) was diagnosed. The epithelium reacted positive for E-cadherin and CK7 by immunohistochemistry (IHC). Bile duct hamartoma is rare in dogs and cats. BDH is reported to be due to failure of involution of embryological remnant during embryogenesis. The current brief report describes cases of bile duct hamartoma in an adult dog and a cat and would contribute to the literature on BDH in felines and canines.

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**Received:** November 22, 2024; **Accepted:** November 25, 2024; **Published:** November 30, 2024

**Keywords:** Bile duct hamartoma, Cat, CK7, Dog, E-Cadherin, Von Meyenburg, Complex

### Introduction

Von Meyenburg complex is reported in humans aged 14 year-to-94-year-old and rarely in animals including a two-day-old, female Belgian calf, and adult dogs and cats [1-5]. It was described as embryonic developmental abnormality due to ductal plate malformation and was reported as a solitary, or multifocal mass as cited by various authors [6, 7]. During embryogenesis, bile ducts originate from two different sources. The intrahepatic bile ducts are derived from liver cells, while the others develop from a primary bile duct source. Many more intrahepatic bile ducts are formed in the embryo than are necessary, and the excess ducts involute. Cysts are thought to arise when ducts from these two sources fail to unite or to involute. This maldevelopment probably causes polycystic disease of the liver, which clinically, does not cause functional impairment [5]. In humans, since the first description there are a few published reports of multi-cystic biliary hamartomas globally [1, 4-11]. Multi-cystic biliary hamartomas (MCBH) can occur at any age, although it is more common in patients aged 30-70 years olds, and two times more likely affect men than women with ratio of 10:6. Because it can occur at a younger age it is important to consider diagnosis of multi-cystic biliary hamartomas in suspected cases in younger age groups. Cystic dilations may not always be the case as was not observed in the bile duct hamartoma reported in a two-day-old female Belgian-Blue calf [5]. Clinically, complaints of some patients include abdominal pain, while others may have no obvious clinical manifestations and cysts are usually discovered by accident. Hence the symptoms may not be typical, and diagnosis

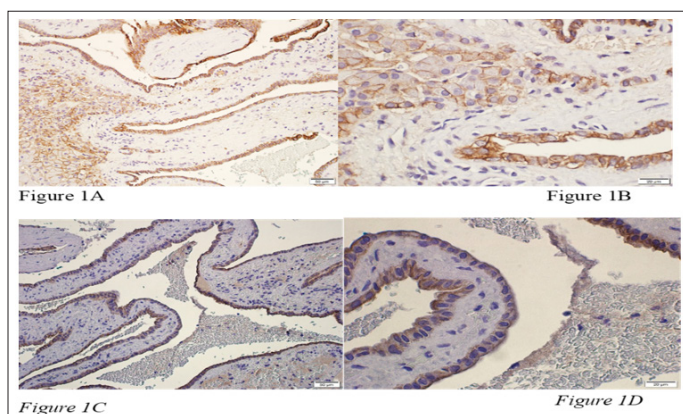
requires a combination of imaging and pathology. Under normal circumstances, the prognosis of MCBH is good. Due to the non-specific radiology, preoperative diagnosis is difficult, and is usually diagnosed by postoperative pathology. Complete resection is the best treatment option, and the postoperative prognosis is good [3].

### Materials and Methods

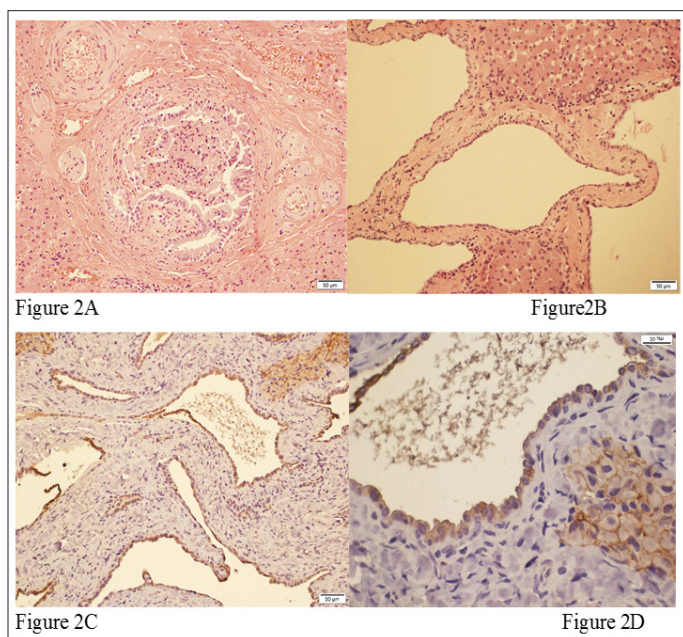
In the current case, biopsies of liver from a 10-year-one-month-old male neutered dog and an adult, male domestic short haired cat of unknown age were received and microscopically examined. Sections were processed for routine histopathology, stained with Hematoxylin Eosin, and examined under light microscopy.

### Results

The examined liver tissues in both the cat and the dog contained aggregates or closely apposed varisized cysts separated by small islands of hepatic parenchyma. The cysts were lined with low simple cuboidal epithelium encompassed with variable amount of fibrovascular tissue. Mitotic cells and malignant features, other significant lesions and evidence of hepatic neoplasia were not observed in both cases. The tissues were subjected to IHC using E-cadherin (1:200 dilution, Mouse IgG2a,  $\kappa$ , BD Transduction Laboratories, BD Biosciences, Franklin Lakes, New Jersey, Cat. # 610182) and CK7 (1:50 dilution, monoclonal, mouse antibody, DAKO, Carpinteria CA, Cat. # M7018) following the manufacturers' procedures and counterstained with hematoxylin. The BDH and adjacent hepatocyte membranes reacted positively for E-cadherin and the bile duct epithelium additionally reacted positively for CK7 (Figure 1 and 2).



**Dog, Figure 1:** Bile Duct Hamartoma and Adjacent Hepatocyte Membranes Reacted Positive for E-cadherin (1A and 1B) and CK7 (1C and 1D) by Immunohistochemistry.



**Cat, Figure 2:** Bile Duct Normal (2A); Bile Duct Hamartoma (2B); Bile Duct Epithelium and Adjacent Hepatocyte Membranes Reacted Positive for E-cadherin (2C) and CK7 (2D) by Immunohistochemistry.

## Discussion

E-cadherin positivity by IHC noted in the hepatocytes and BDH in the dog in this report suggest the intrahepatic BDH in the dog in the current case likely developed from hepatocytes and not from bile duct source as described earlier<sup>5</sup>. Loss of E-cadherin especially in the intrahepatic bile duct epithelium functionally impair biliary flow and subsequently induce cholestatic liver injury and sclerosing cholangitis [8]. Loss of E-cadherin was not observed in both the cat and the dog liver in this report. Generally, bile duct hamartoma is a developmental abnormality, often multifocal, not diffuse, diagnosed in animals and humans at very young to adult ages, it is an incidental finding, and not a life-threatening condition.

Owing that it is reported in adult humans and dogs and cats and the fact that it is multifocal or solitary mass and not involving the entire bile ducts throughout the liver of affected patients it possibly developed due to any local or multifocal effect of an instigating insult to already developed bile duct in adults. This

needs further investigation. The issue of overgrowth of cells due to growth-abnormalities-triggered changes later in life after normal development of the ducts could underlie the BDH in adults rather than congenital malformation that emanated at birth. Generally, it appears plausible to consider that not all bile duct hamartomas are congenital; and often it is reported as idiopathic or accidental finding without clinical impact. The current report would further contribute to the available literature and understanding of bile duct hamartoma in cats and dogs.

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