CASE REPORT

A case of a congenital umbilicobiliary fistula associated with gallbladder agenesis in a dog

Um caso de uma fistula umbilicobiliar associada com agenesia da vesícula biliar num cão

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Abstract

Congenital biliary tree malformations are rarely described in the veterinary field. A congenital umbilicobiliary fistula associated with gallbladder agenesis was reported here for the first time in a 1-year-old male French bulldog. After contrast study, abdominal ultrasound, and histopathology, we concluded that the umbilicobiliary fistula was an aberrant duct that originated directly from the expected location of the cystic duct and gallbladder. The clinical case was treated surgically through ligation and excision of the aberrant duct before entering the common bile duct. The recovery and long-term follow-up were uneventful. The pathophysiology of biliary congenital malformations is discussed, along with clinical considerations that should be considered in similar future cases.

Keywords: biliary tract diseases, aberrant biliary duct, gallbladder atresia, congenital abnormalities, dogs.

Resumo

As malformações congênitas da árvore biliar são raramente descritas na veterinária. Uma fístula umbilicobiliar congênita associada à agenesia da vesícula biliar foi relatada aqui pela primeira vez em um buldogue francês macho de 1 ano de idade. Após estudo contrastado, ultrassonografia abdominal e histopatologia, concluímos que a fístula umbilicobiliar era um ducto aberrante que se originava diretamente da localização esperada do ducto cístico e da vesícula biliar. O caso clínico foi tratado cirurgicamente através da ligadura e excisão do ducto aberrante antes de entrar no ducto colédoco. A recuperação e o acompanhamento a longo prazo transcorreram sem intercorrências. A fisiopatologia das malformações congênitas biliares é discutida, juntamente com considerações clínicas que devem ser consideradas em casos futuros semelhantes.

Palavras-chave: doenças do trato biliar, ducto biliar aberrante, atresia da vesícula biliar, anomalias congênitas, cães.

Introduction

Developmental biliary ductal anomalies include cysts, aberrant ducts, and atresia (Ando, 2010; Center, 2011; Sato et al., 2018). Biliary fistulae are most frequently acquired, but congenital fistulae can occur in the gastrointestinal or respiratory tracts (Sachdev et al., 2011). Most acquired fistulas can be cholecystocutaneous, cholecystoenteric, or between biliary ducts and the gastrointestinal or respiratory tracts (Crespi et al., 2016). Congenital umbilicobiliary fistulas were reported twice in veterinary and medical literature, one case in a 1-year-old male English bulldog and the other in a 10-day-old girl. Both cases seemed very similar, with one biliary fistula associated with a normal gallbladder (Mohta et al., 2006; Sides et al., 2004).

In a retrospective study of 17 dogs with gallbladder agenesis, concurrent vascular and biliary duct anomalies were observed, but none of the concurrent anomalies were an umbilicobiliary fistula (Sato et al., 2018). In humans, one-third of the patients with gallbladder agenesis have associated bile duct atresia or other multiple local or distant congenital anomalies, which are considered rare congenital biliary fistulas (Günlemez et al., 2009; Sachdev et al., 2011). Therefore, to our knowledge, synchronous congenital gallbladder agenesis and umbilicobilliary fistula has not been reported in the veterinary field. The aim of this case report is to describe the clinical

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and anatomical features and management of umbilicobilliary fistulas, followed by a review of the current literature.

Case report

A 1 year-old, 11.9 kg, male, French bulldog had a history of yellow umbilical discharge since birth and did not have any other clinical signs, including normal growth and development, and no abnormal micturition when the owner was inquired.

Upon inspection, the umbilical orifice was open. A low volume of yellow discharge flowing at a very slow rate was identified from this orifice. The periumbilical skin was normal and there was no associated herniation. No abdominal pain was noted on palpation; however, abdominal cryptorchidism was found. No other relevant anomalies were identified on clinical examination.

Routine blood work was not performed due to the owner's economic restrictions, but ultrasound and umbilical liquid analyses were conducted. Umbilical liquid biochemical analysis revealed biliary acids, and ultrasound examination was able to follow the duct that led to the common bile duct. There was an absence of the gallbladder and there was no intrahepatic or extrahepatic duct dilation.

Surgery was planned, which included a fistulogram prior to the intervention. The animal was premedicated with a combination of midazolam (0.2mg/kg) and fentanyl (3μg/kg), which were administered intravenously after placement of a 23G intravenous catheter in the right cephalic vein. Intraoperative fluid therapy was administered with a saline solution at a dose of 5mL/kg/hour throughout the procedure. Pre-oxygenation was initiated with an oxygen mask until induction, which was performed intravenously using 2mg/kg of propofol. After intubation, maintenance was ensured with isoflurane (0.5-1.0%) and oxygen at 30mL/min. The heart rate, respiratory rate, mucus membrane color, capillary refill time, indirect blood pressure, PaO_2, $ETCO_2$, ECG, and body temperature were monitored. The patient was warmed using a blanket and a warm rubber water bottle.

Fistulography was performed by gently injecting a 2mL/kg iohexol 300mg/mL contrast solution (Omnipaque, GE Healthcare A.S.) through a 24G intravenous catheter inserted into the umbilical opening. In lateral and ventrodorsal radiography, direct communication from the umbilical opening to the common bile duct was identified. Contrast medium extended to the left and right intrahepatic biliary channels and extrahepatic biliary system, confirming the absence of the gallbladder. In addition, contrast medium was identified in the gastrointestinal tract (Figure 1).

Differential diagnoses included a patent vitellointestinal duct, cutaneous biliary fistula, and aberrant extrahepatic malformation.

Exploratory laparotomy has been proposed as the definitive diagnostic and treatment procedure. Intraoperative analgesia was assured with the administration of fentanyl at a previously described dosage, repeated every 30 minutes. Meloxicam was administered intraoperatively to manage postoperative pain. After cutaneous access, dissection was performed around the umbilical orifice. A *linea alba* incision was made to explore the posterior aspect of the abdominal wall and the trajectory of a possible duct or fistula. After identification of a duct emerging from the common bile and/or cystic duct, it was dissected to the abdominal wall through the falciform ligament until it connected with the skin (Figure 2). As expected, there was no gallbladder and the persistent duct was communicating at the level of the expected location of the cystic duct. Ligation was performed at the opening site of the common bile duct. At this level, it was possible to identify a small artery, closely associated with the dorsal aspect of the aberrant duct, at the expected location of the cystic artery and ligated along with the excised duct. Abdominal access was closed routinely.

Histopathological analysis of the excised tubular structure revealed histological features identical to those of the gallbladder and other major bile ducts. Therefore, this tubule corresponds to a congenital malformation, that is, an aberrant bile duct.

Postoperative recovery was uneventful, without any related complications during the 2 year follow up.

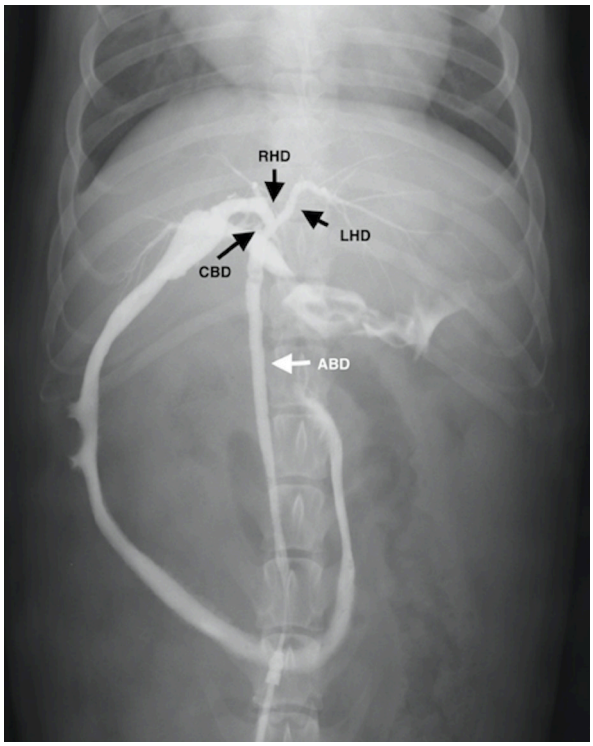

Figure 1. Fistulogram through the umbilical fistula showing communication with the common bile duct (CBD), ventro-dorsal view, in a 1-year-old male French Bulldog. It is possible to observe the aberrant bile duct (ABD) meeting the CBD, after the left and right hepatic duct (LHD and RHD, respectively) gathering. The spread of contrast to the gastro-intestinal tract (stomach, duodenum, and jejunum) can also be observed.

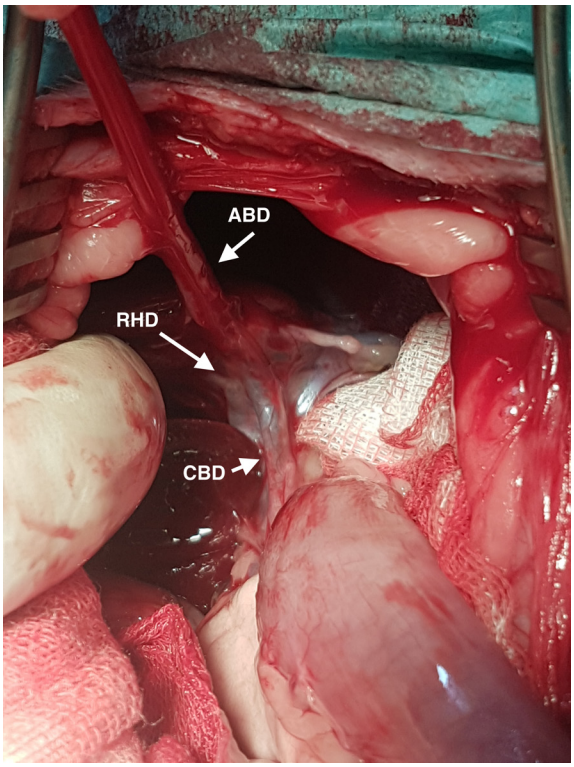

Figure 2. Intraoperative image identifying the aberrant duct (ABD) after dissection from the falciform ligament, the right hepatic duct (RHD), and the common bile duct (CBD).

Discussion

Abnormal bile duct and gallbladder agenesis can result from differentiation defects and abnormal duct development. Failure to vacuolize the caudal portion of the hepatic diverticulum, the *pars cistica*, is thought to be the cause of an absent gallbladder, a rare anomaly that may or may not be associated with clinical signs (Kelly et al., 2019; Sato et al., 2018). It can occur alone or in association with other biliary developmental anomalies, such as ductal plate malformations (Pillai et al., 2016; Sato et al., 2018).

The umbilical cord is formed after the enveloping of the amnion tissue from the body stalk, omphalomesenteric duct, and umbilical coelom, around 2 weeks of gestation in dogs (Aralla et al., 2013). Primitive umbilical cord formation precedes hepatic diverticulum differentiation, but there is an intimate proximity between these two structures at the level of the falciform ligament. The peritoneal ligament attaches the liver to the front body wall and separates the left and right lobes. The umbilical vein runs through the falciform ligament ventrally into the umbilical cord, which, after birth, leads to the round ligament of the liver and ventral part of the falciform ligament. Within the umbilical cord, the umbilical vein, vitelline duct (the previous omphalomesenteric duct), allantois duct, and umbilical arteries were enrolled by Wharton's jelly and amnion after the body wall. In the case reported here, this anomaly could have been a result of excessive ventral invagination with entrapment of the cystic diverticulum, resulting in an aberrant herniation that, after birth, gave rise to direct communication to the exterior. This explanation is supported by the histological and anatomical features observed: absence of the gallbladder and a histologically confirmed bile duct from the umbilicus to the common bile duct at the expected location of the cystic duct.

The differential diagnosis in this case was the persistence of the vitelline duct or the urachus, and rare umbilical developmental anomalies, discarded after umbilical discharge analysis, ultrasound, and contrast study (Peterson, 2011). Fistulography was useful to evaluate anomalies in relation to the rest of the hepatic tree and to assist in the decision to perform surgical correction. This strategy was similar to that previously reported (Sides et al., 2004). Computed tomographic cholangiography would provide more detailed information (Kelly et al., 2019; Sato et al., 2018); however, it was not performed for economic reasons. Regardless, the surgical approach was expected to be straightforward, resulting in the correction of a persistent biliary discharge that could lead to an ascending liver infection. In this case, the fistula extended from the umbilical orifice to the common hepatic duct, and in the previous case, the communication was with the right hepatic duct (Sides et al., 2004).

Conclusions

The interest in this exceptional case relies on the importance of considering such congenital anomalies as possible biliary anomalies that are compatible with life and surgically correctable if no major changes arise from the extrahepatic or intrahepatic biliary system. Moreover, surgical correction must be performed, otherwise, an increased risk of bacterial hepatitis of a cutaneous origin can occur.

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Ethics statement

Those responsible for the animal formally consented to the study.

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Conflict of interests

No conflict of interests declared concerning the publication of this article.

Authors' contributions

All authors contributed equally to the handling of the case report and writing of the manuscript.

Availability of complementary results

Any complementary information is available from the authors on request.

The study was carried out at VetAlmada in collaboration with the Faculty of Veterinary Medicine, Universidade de Lisboa, Portugal.

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