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Intrathoracic Ectopic Liver in a Dog: A Case Report

Mohammad Naser Nazem[1*], Sayed Mohsen Sajjadian[1], Bahador Shojaei[1], Reza Kheirandish[2], Mohammad Hasanzadeh[1]

1. *Department of Basic Sciences, Faculty of Veterinary Medicine, Shahid Bahonar University of Kerman, Kerman, Iran*
2. *Department of Pathobiology, Faculty of Veterinary Medicine, Shahid Bahonar University of Kerman, Kerman, Iran*

Abstract

The accessory hepatic lobe is an extremely rare anomaly reported in humans and animals. A large domed solid mass and two smaller masses were noticed during thorax dissection in a six-year-old mixed breed male dog with no previous history of the disease, which was fixed to be used in the anatomy hall. The masses were placed adjacent to the diaphragm, between the lung's right and left caudal lobs. The masses with a common thick vascular pedicle had pierced the diaphragm and run to the falciform ligament of the liver. Histological findings showed liver tissue and hepatocytes were arranged radially around the central vein. There were sinusoids between the hepatocyte plates, dilated as telangiectasia in some areas. The study of the pedicle revealed a normal elastic artery, normal vein, and normal biliary duct crossed to the falciform ligament in the abdomen.

The macroscopic and microscopic findings revealed type I intrathoracic ectopic liver.

KEYWORDS: Diaphragm, Dog, Intrathoracic, Liver, Pedicle

Correspondence

Mohammad Naser Nazem, Department of Basic Sciences, School of Veterinary Medicine, Shahid Bahonar University of Kerman, Kerman, Iran. Tel: +98 (034) 31322954, Fax: +98 (034) 33222047, Email: nnazem@uk.ac.ir nasernazem@yahoo.com

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Case History

The accessory hepatic lobe is an extremely rare anomaly (Smiley *et al.*, 2013; An *et al.*, 2010; Massaro *et al*., 2007; Shrestha *et al.*, 2014; Chen *et al.*, 2014), reported in human beings (An *et al*., 2010; Jambhekar *et al*., 2010; Smiley *et al*., 2013; Chen *et al.*, 2014) and animals (Machado & Lozzio, 1972; Dhaliwal & Lacey, 2009; Hifumi *et al.*, 2015; Hifumi *et al*., 2014). The unusual presence of liver tissue has been commonly described in some organs, including the gall bladder, umbilical fossa, adrenal gland, pancreas, and the spleen (An *et al*., 2010; Jambhekar, *et al*. 2010; Mehta *et al.*, 2010; Choi *et al*., 2008). An accessory lobe of the liver is a rare congenital anomaly, which can undergo torsion and be presented as an acute surgical emergency (Jambhekar *et al.*, 2010).

There have been reports of a supradiaphragmatic ectopic liver in the chest cavity and pericardium (Iber & Rintala, 1999; Yucel *et al.*, 2015). In most cases, the supradiaphragmatic ectopic liver was accompanied by a transdiaphragmatic pedicle in the main body of the liver within the abdominal cavity (Iber & Rintala, 1999; Yucel *et al.*, 2015). In some other cases, the supradiaphragmatic ectopic liver was utterly separated from the abdominal cavity without connecting the thorax and the abdomen, and diaphragmatic anomalies (Chen *et al.*, 2014; Mendoza *et al.*, 1986).

An accessory or intrathoracic ectopic liver lobe is often asymptomatic clinically (Iwaki *et al.*, 2017). Trauma, postoperative complications, congenital diaphragmatic hernias, or other malignancies affecting the diaphragm are the more typical circumstances where the liver tissue is found in the thoracic cavity (Iber & Rintala, 1999; Yaguchi *et al.*, 2015; Iwaki *et al.*, 2017). It should be noted that this malformation has no sex predilection (Vercelli-Retta, 1978).

Ultrasonography or CT-guided biopsy (Choi *et al.* 2008) and MRI (Jambhekar *et al.* 2010) may contribute to establishing an accurate diagnosis. In one case, the supradiaphragmatic heterotopic liver was misdiagnosed as pulmonary sequestration. Color Doppler ultrasonography or angiography may show a feeding vessel and also contribute to differentiating pulmonary sequestrations from the heterotopic liver (Lee *et al.*, 2016). The diagnosis is usually made after laparotomy by histopathological confirmation (Jambhekar *et al.*, 2010).

In veterinary medicine, intrathoracic ectopic liver induced by traumatic injury has been reported in cats (Dhaliwal & Lacey, 2009), cows (Hifumi *et al.*, 2014), and dogs (Hifumi *et al.*, 2015). This study describes an intrathoracic ectopic liver, which might have been induced by a congenital abnormality in a dog.

Clinical Presentation

A 6-year-old mixed breed male dog weighing 15 kg was fixed by formalin to be used as a model in the anatomy hall of the Anatomy Department of the Veterinary Faculty at the Shahid Bahonar University of Kerman, Iran. There was no previous history of traumatic diaphragmatic hernia or surgery, and no clinical abnormalities were noticed before its submission for fixation.

During the thorax dissection, the gross examination revealed a large domed solid mass, approximately 12.5×5 cm at its base and height, respectively. Moreover, two significantly smaller roundish masses were positioned ventral; the larger one was about 3.5×1.5×1.2 cm (Figures 1 & 2).

The base of the larger mass and the two smaller ones were placed near the diaphragm, towards the left side of the thorax, between the right and left caudal lobs of the lung. The accessory lobe of the lung was placed at the left side of the larger mass (Figure 3). All masses with a common thick vascular pedicle had pierced the diaphragm and run to the liver's falciform ligament (Figure 1). All masses were brown and capsulated (except around the pedicle) with an irregular surface and had a firm consistency such as liver that differentiates it from the lung (Figures 1 and 2).

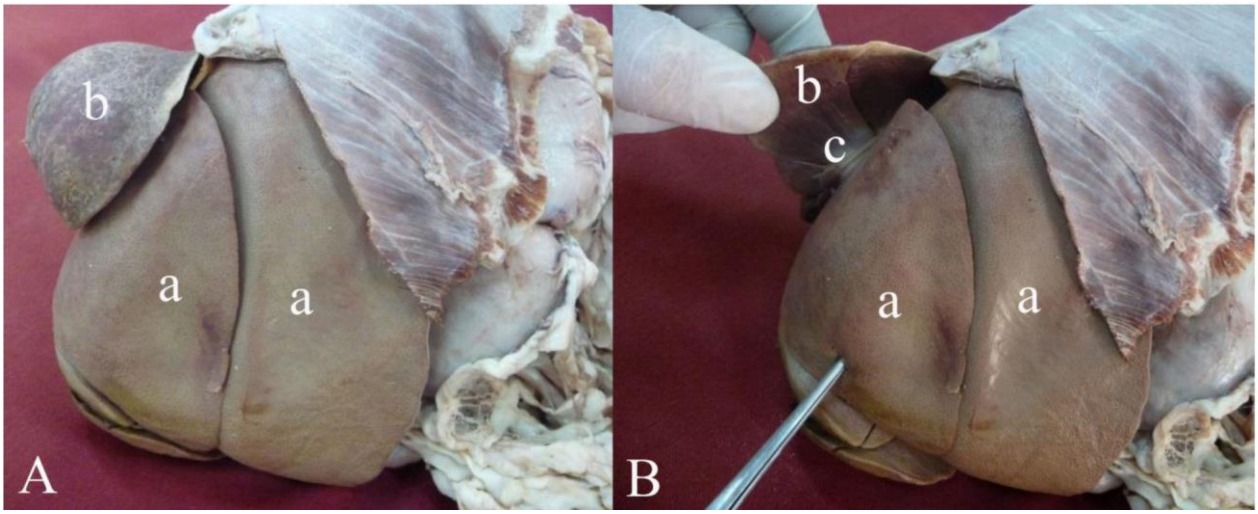

Figure 1. Gross cut surface of the formalin-fixed liver. A) Liver (a), Accessory liver (b). B) Liver (a), Accessory liver (b), and its pedicle (c).

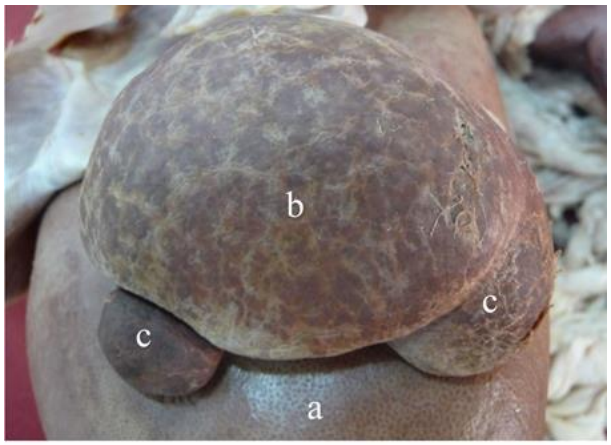

Figure 2. Gross appearance of the liver (a), main mass (b), and two smaller masses (c) of the accessory. Cranial view.

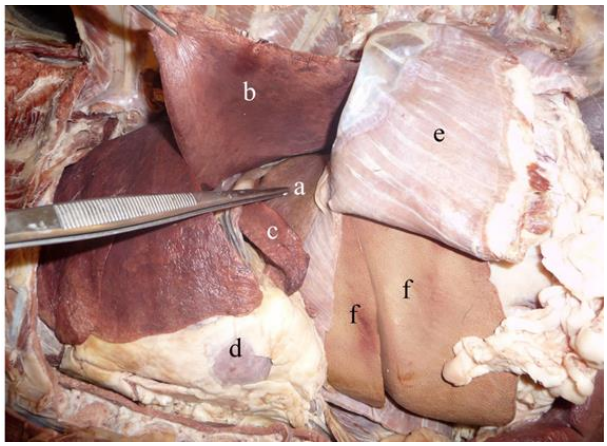

Figure 3. The position of the supradiaphragmatic accessory liver in the thoracic cavity. A) Accessory liver, b) Caudal lobe of the left lung, c) Accessory lobe of lung, d) Heart, e) Diaphragm, f) Liver.

Diagnostic Testing

To perform histopathological evaluation, 1×1×1 cm samples were taken from the three different parts of the masses, washed with PBS (pH 7), and placed in a 10% buffered formalin solution (pH 7.2). After ten days, the samples embedded in paraffin were fixed in a 10% neutral formaldehyde solution and dehydrated in ethanol and xylol alcohols. The 5 µm sections were stained with the H&E technique for evaluation with a light microscope.

The mass revealed histological features consistent with the liver tissue in the microscopic examination. Hepatocytes were polygonal with the central nuclei and arranged radially around the central vein. There were sinusoids between the hepatocyte plates, which were dilated as telangiectasia in some areas. The portal areas consisting of interlobular bile ducts, interlobular arteries, and interlobular veins were also present. The central veins were also noticed at the center of the hepatic lobules (Figures 4, 5, & 6). The study of the pedicle revealed a normal elastic artery, normal vein, and normal biliary duct crossed to the falciform ligament in the abdomen. Moreover, the intrathoracic ectopic liver was diagnosed according to the histological findings. However, the histopathologic study revealed no hepatic fibrosis or vascular structure disorders. No gross abnormalities were evident in the other organs.

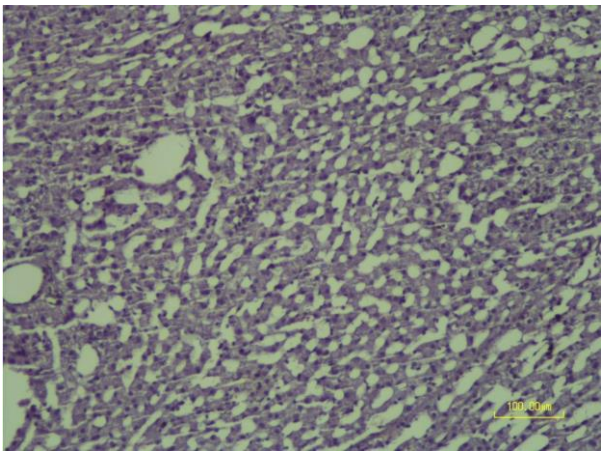

Figure 4. Polygonal hepatocytes arrange radially around the central vein, and there are sinusoids between them (H&E, Bar=100 µm).

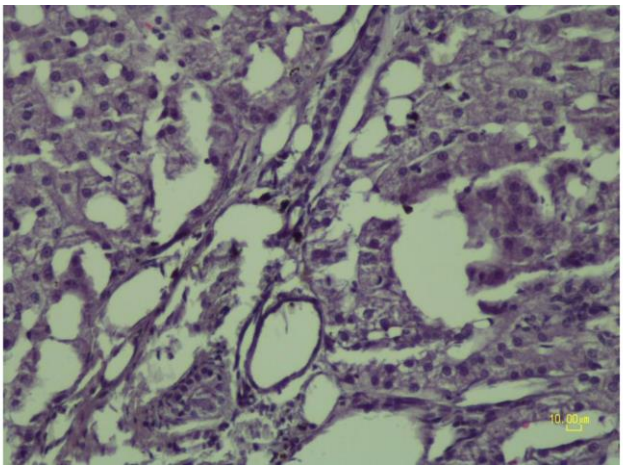

Figure 5. In this micrograph, some bile ducts were seen (H&E, Bar=10 µm).

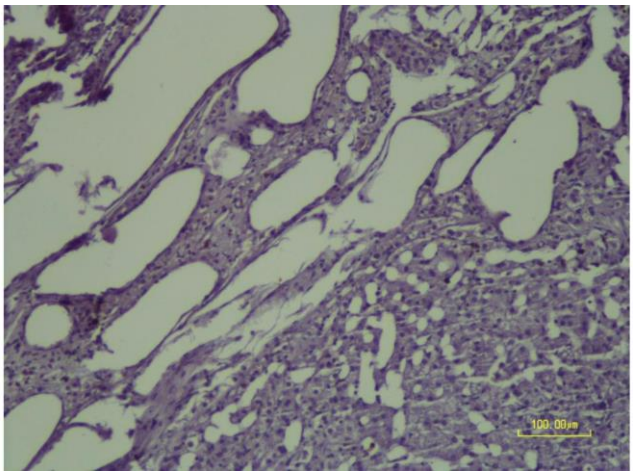

Figure 6. This micrograph shows telangiectasia (H&E, Bar=100 µm).

Assessments

In humans, ectopic liver masses are sometimes found in the abdominal cavity, especially on the surface of the gall bladder and in the hepatic ligament (Chen *et al.*, 2014). As mentioned before, the supradiaphragmatic ectopic liver is a rare anomaly in both humans and animals (Dhahwal & Lacey, 2009; An *et al.*, 2010; Jambhekar *et al.*, 2010; Smiley *et al.*, 2013; Chen *et al.*, 2014; Hifumi *et al.*, 2014; Hifumi *et al.*, 2015; Ito *et al.*, 2016; Sag *et al.*, 2018;). Adult patients usually exhibit no specific symptoms (An *et al.* 2010; Ito *et al.* 2016); however, there are a few reported cases with clinical symptoms. The most common symptoms in pediatric patients are respiratory distress and chest pain (An *et al.*, 2010). In one case, the patient exhibited thoracic pain induced by an incomplete torsion of an intrathoracic ectopic liver lobe (Sehdeva & Logan, 1971). Moreover, mild asymmetry of the chest (Iber & Rintala, 1999), cough and fever (Choi *et al.*, 2008), dyspnea (An *et al.*, 2010), and abdominal pain (Massaro *et al.*, 2007; Jambhekar *et al.*, 2010; Ito *et al.*, 2016) are also reported in some cases.

The ectopic liver has been classified into four types (Arslan *et al.* 2014): 1) The accessory lobe of the liver with a considerable size and a connecting stalk to the liver, 2) a small accessory lobe of the liver attached to the liver, (3) ectopic liver with no connection to the liver, and (4) the microscopic ectopic liver tissue. The first or second types of ectopic liver mainly occur in the intraabdominal cavity;

however, few cases of supradiaphragmatic ectopic are reported in the literature. The case described in the present study belongs to the first type. Although the most probable cause of this event is the formation of an accessory liver lobule with regression or atrophy of the original connection to the true liver (Mendoza *et al.* 1986), we presented an intrathoracic accessory liver lobe with a pedicle connected to the abdominal liver. The case was similar to those in some studies describing a mass attached to the liver proper by a vascular pedicle, all of which are mainly transdiaphragmatic (Chen *et al.* 2014; Yucel *et al.* 2015).

Three typical sites are described for this anomaly, with the right costophrenic variety being the most common site (Vercelli-Retta, 1978) in humans (An *et al.*, 2010; Jambhekar *et al.*, 2010) and animals (Hifumi *et al.*, 2015; Machado and Lozzio, 1972). The other sites are the left supradiaphragmatic liver lobe and adjacent to the inferior vena cava (Vercelli-Retta, 1978). In our case, the supradiaphragmatic mass was located at the left side of the hemithorax, close to the heart. This finding was consistent with those reported in previous studies (Hifumi *et al.* 2014; Yucel *et al.* 2015).

Classification can also be based on the biliary drainage and the presence or absence of a common capsule (Type I). The separate accessory lobe duct drains into an intrahepatic bile duct of the normal liver (Type II), and the separate accessory lobe duct drains into an extrahepatic bile duct of the normal liver (Type III). The accessory lobe and the normal liver have a common capsule, and the bile duct of the accessory lobe drains into an extrahepatic duct (Arslan *et al.*, 2014). In our study, the separate accessory lobe duct drainage into an intrahepatic duct was observed (Type I). There are intralobar and extralobar forms embedded in the normal lung, while the latter is separated from the adjacent lung by its visceral pleural investment (Kim *et al.*, 2016). In our report, we described an intralobar form of the intrathoracic liver because of the accessory liver lobe between the lung's right and left caudal lobes.

On the other hand, the lack of bile accumulation in the liver sections suggests the likelihood of communication between the ectopic pedicle and the biliary tree of the abdominal liver. The microscopic study may propose that this communication was via the falciform ligament. In other words, the histological analysis revealed no evidence of cirrhosis or cholestasis (Figures 4, 5, & 6). It showed appropriate functional biliary drainage of accessory lobe to the vascular supply within the pedicle.

According to Machado and Lozzio (1972), the ectopic hepatic lob shows hepatocytes positioned in the uncommon rows of variable thickness surrounding abnormally dilated sinusoids. Furthermore, severe congestion, ruptured sinusoids, focal hemorrhage, clusters of blood cells, and bile pigment in the areas of hemorrhage were also observed. None of these abnormalities were noticed in the present study; however, there was a three-part intrathoracic liver with no completely-separated parts.

In some animals, the supradiaphragmatic acessory lobe is reported along with other anomalies such as hydronephrosis (Machado and Lozzio, 1972), which were not noticed in our case.

The possible heritability of the accessory lobe of the liver is unknown (Machado & Lozzio, 1972). According to the literature, several possible mechanisms have been proposed for the development of the heterotopic liver. In pediatric and young adult patients presenting supradiaphragmatic liver with a connection such as a transdiaphragmatic pedicle into the liver proper and/or diaphragmatic hernia, it can be explained by an anomaly in the development of diaphragm and liver bud (Lande *et al.*, 2015).

Embryologically, the hepatic bud originated from the foregut grows into the mesenchyme of the septum transversum during the fourth week of gestation and proliferates actively (Chandramohan *et al.*, 2014). At approximately the same time, the diaphragm develops centrally from the septum transversum and peripherally from the right and left pleuroperitoneal membranes (Shrestha *et al.*, 2014). Normally the pleuroperitoneal cavity is closed during the sixth and seventh week of gestation. A small portion of proliferating hepatic tissues may grow into the thoracic cavity before the complete closure of the diaphragmatic membrane, and the defect in the fusion of the diaphragmatic membranes may allow the sequestration of the hepatic tissue. Accordingly, the supradiaphragmatic liver appears with regression or atrophy of connection to the abdominal liver. Another possibility could be the development of an

entirely different liver bud independent of the main hepatic diverticulum with no prior connection (Mendoza *et al.* 1986). This mechanism might explain the pathogenesis of the supradiaphragmatic liver with no pedicle into the liver proper.

Another explainable mechanism for the minority of cases reported in the literature was associated with previous trauma history (An *et al.*, 2010). According to some reports, the most plausible explanation was that the fragment of the liver parenchyma was introduced into the thoracic cavity during the previous trauma, followed by regeneration and replace-ment of a nodular mass (Chen *et al.*, 2014). This mechanism seems to be more likely in middle-aged or old patients with no diaphragm and liver abnormalities in their childhood and young adulthood, who underwent later traumatic injury on the lower chest or/and upper abdomen. We failed to detect the reason for the presence of this abnormality in our case.

Based on our researches, this report describes an intrathoracic ectopic liver, which might have been induced by a congenital abnormality in the dog.

Conclusion

There are few case reports of an intrathoracic ectopic liver in veterinary medicine. To our knowledge, this is one of the rare incidence of this anomaly in dogs. This anomaly has not been recognized as a severe clinical problem.

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Conflict of Interest

The authors declared no conflict of interest.

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مجله طب دامی ایران، ۱۴۰۰، دوره ۱۶، شماره ۲، ۲۰۲-۲۰۹

کبد نابجای داخل سینه‌ای در یک قلاده سگ: گزارش موردی

محمدناصر ناظم*[۱]، سید محسن سجادیان[۱]، بهادر شجاعی[۱]، رضا خیراندیش[۲]، محمد حسن زاده[۱]

[۱] گروه علوم پایه، دانشکده دامپزشکی دانشگاه شهید باهنر کرمان، کرمان، ایران

[۲] گروه پاتوبیولوژی، دانشکده دامپزشکی دانشگاه شهید باهنر کرمان، کرمان، ایران

(دریافت مقاله: ۲۰ مهر ماه ۱۴۰۰، پذیرش نهایی: ۰۵ دی ۱۴۰۰)

کبد اضافی، یک ناهنجاری بسیار نادر، در انسان و حیوانات، گزارش شده است. طی تشریح قفسه سینه یک قلاده سگ نر ۶ ساله نژاد مخلوط فیکس‌شده برای سالن تشریح که هیچ سابقه‌ای از بیماری نداشت، یک تودهٔ بزرگ همراه با دو تودهٔ کوچکتر دیده شد. تمام توده‌ها، نزدیک دیافراگم و بین لوب راست و چپ ریه قرار داشتند. توده‌ها، دارای یک پایک ضخیم مشترک بودند که دیافراگم را سوراخ کرده و همراه با لیگامان فلسی شکل کبد، ادامه می‌یافت. یافته‌های هیستوپاتولوژی، بافت کبدی را نشان داد. هپاتوسیت‌ها به صورت شعاعی، اطراف ورید مرکزی حضور داشتند. سینوزوئیدها که بین صفحات هپاتوسیت‌ها مشاهده می‌شدند، در برخی نواحی، اتساع یافته بودند. مطالعه پایک، یک سرخرگ الاستیک، سیاهرگ و مجرای صفراوی طبیعی را نشان داد که در لبه لیگامنت فلسی شکل، به شکم وارد می‌شدند. بر اساس یافته‌های ماکرسکوپیک و میکروسکوپیک، عارضهٔ فوق، کبد نابجای داخل سینه‌ای تشخیص داده شد.

واژه‌های کلیدی: کبد، داخل سینه‌ای، پایک، دیافراگم، سگ

نویسندهٔ مسئول: محمدناصر ناظم، گروه علوم پایه، دانشکده دامپزشکی دانشگاه شهید باهنر کرمان، کرمان، ایران ایمیل nasernazem@yahoo.com nnazem@uk.ac.ir