GALL-BLADDER AND HEPATODUODENAL LIGAMENT LYMPHANGIOMA – CASE REPORT AND LITERATURE REVIEW

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Lymphangiomas are rare benign lesions of the lymphatic vessels that are most commonly diagnosed in childhood. Intraperitoneal localization is unusual as, typically, they are located in the head and neck areas. In general, abdominal lymphangiomas seem to be asymptomatic, however, patients may occasionally suffer from acute abdominal symptoms, due to intestinal obstruction or peritonitis. The study presented a case of a 41-year-old female patient, clinically asymptomatic, who was accidentally diagnosed with a multiseptated cystic lesion of the right liver lobe surrounding the gall-bladder fossa in a routine ultrasound examination. Further examinations including computed tomography and magnetic resonance (MR) aroused suspicion of a polycystic lesion of the gall-bladder and hepatoduodenal ligament. The cystic lesion of the gall-bladder and hepatoduodenal ligament filled with lymphatic fluid was diagnosed intraoperatively. Simultaneous cholecystectomy and radical resection of the cystic lesion was undertaken. The histopathological examination revealed the presence of a lymphangioma. Additionally, the authors of the study reviewed literature data concerning gall-bladder lymphangiomas.

Key words: lymphangioma, gall-bladder, cholecystectomy, lymphatic vessels

Lymphangiomas are rare lymphatic vessel malformations (1 in 2-4 thousand living births) most commonly diagnosed during childhood, being located in the head and neck areas (1-7). Only 5% of them relate to the abdominal cavity, most commonly to the small and large bowel, mesentery, omentum, retroperitoneal space, gall-bladder, and pancreas (3, 4, 5, 7-10). Approximately 60% of these cases are diagnosed before the age of 5 years. However, many remain clinically asymptomatic until adulthood (3, 5). The rare occurrence of the above-mentioned directly translates to the small number of publications concerning the imaging of these lesions and thus, diagnostic problems (11, 12, 13). Often, proper diagnosis is accidentally determined during routine examinations, including surgery and histopathological examinations (1, 4). The study presented a case of a female patient with gall-bladder and hepatoduodenal ligament lymphangioma, as well as literature review concerning the issue.

CASE REPORT

A 41-year old female patient was admitted to the Department with suspicion of a liver hilus cyst, accidentally diagnosed four years previously during abdominal ultrasound. The cyst was described as a multilocular lesion located in the right hepatic lobe, 93 x 42 mm in size, without signs of flow. The next ultra-
sound examination performed four years later showed the presence of a lesion, 146 x 64 mm in size (fig. 1). The patient was directed for further diagnosis. Computed tomography confirmed the presence of a cystic lesion with numerous multilocular connective tissue septates, 13 x 5.5 x 6.5 cm in size, located in the hilus of the liver, and not the right lobe, modeling (without infiltration) and displacing the gall-bladder and cystic duct, medially and inferiorly. The ELISA examination for the presence of IgG antibodies against hydatid cyst antigens proved negative (0.092, with the threshold value at 0.715).

On admission, the patient was asymptomatic. She remained under hematological control, due to recurrent iron deficiency anemia. The physical, hematological, and biochemical examinations showed no abnormalities. The tumor markers (AFP, CEA, CA 19-9) were within normal limits. The patient was subject to repeated computed tomography and magnetic resonance imaging examinations (fig. 2 and 3), which demonstrated that the cystic lesion previously described showed discreet signs of contrast enhancement, modeling the gall-bladder, cystic and common hepatic ducts. The above-mentioned image was unclear, however, enabled to exclude a biliary pathology. After preparation the patient was subject to surgical intervention. During surgery we observed a thin-walled gall-bladder surround-

![Fig. 1. Abdominal ultrasound showing the multilocular lesion located in the right hepatic lobe, 146 x 64 mm in size](image1)

![Fig. 2. Computed tomography in the sagittal (a) and frontal (b) planes, showing the cystic lesion with numerous connective tissue septa, 13 x 5.5 x 6.5 cm in size, located in the hilus of the liver, modeling (without infiltration) the gall-bladder and cystic duct](image2)
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Tomical variation of the common hepatic duct, which was created from the combination of four intrahepatic ducts (fig. 5). Afterwards the patient was subject to cholecystectomy with radical resection of the lesion (fig. 6). Both the lesion and its contents were subject to histopathological evaluation. The other abdominal cavity organs were macroscopically normal. The histopathological result was as follows: thin-walled, non-inflammatory gallbladder, and a thin-walled multilocular lesion corresponding to a lymphangioma. The biochemical evaluation of the fluid content showed features of blood plasma (protein – 5.3 g/dl, glucose – 107mg/dl, bilirubin – 0.3 mg/dl, LDH – 276 U/l). Control ultrasound showed no disease recurrence.

DISCUSSION

Lymphangiomas are a group of benign tumors originating from lymph vessels located in the neck (75%) and armpits (25%). Abdominal cavity hemangiomas constitute 5% of all cases and concern the small and large bowels, mesentery, omentum, retroperitoneal space, gall-bladder, pancreas, liver and falciform ligament (3, 4, 5, 7-10). Although most of the lesions clinically manifest symptoms during childhood, still a significant percentage of patients are diagnosed with the above-mentioned
disease in adulthood, during pre- and intraoperative examinations (3, 5).

The etiology of lymphangiomas is poorly known. It is suggested that an important role is attributed to inflammation and fibrosis, genetic factors, mechanical lymph ducts damage, lymph nodes atrophy, and lymphatic endothelial disturbances (3). Lymphangiomas may be divided into 3 major histological types: (1) simple or capillary hemangioma, (2) cavernous hemangioma, and (3) cystic angioma or cystic hygroma (1, 3).

The disease during the course of lymphangioma is unspecific. In 40% of cases patients are asymptomatic, the hemangioma being incidentally diagnosed during imaging examinations.

Symptoms of an abdominal cavity hemangioma are attributed to the enlarging “cystic hemangioma structures” on surrounding anatomical structures. The most common symptoms include transient pain, nausea, vomiting, abdominal distention, ascites, and fever. Complications of enlarging cysts include inflammation, intestinal or ureter obstruction, cystic rupture or torsion with bleeding, responsible for “acute abdomen” symptoms (1, 4, 7, 14, 15).

As previously mentioned diagnosis is often established during exploratory laparotomy or autopsy. Typical laboratory investigations do not apply in the diagnostics of lymphangiomas. Correct diagnosis is only possible on the basis of imaging examinations (ultrasound, CT, MR). It seems that these examinations complement each other and the use of only one during diagnostics might not be beneficial (4, 7, 11, 12).

In a typical ultrasound image one may observe a multilocular lesion filled with an anechoic content. The internal structures are well visible, including the septa. Computed tomography showed the presence of cystic, multilocular lesions filled with fluid content. After contrast administration the cystic walls were subject to signal enhancement. Unfortunately, hemangioma bleeding or inflammation might not distinguish the above-mentioned lesions from cystic tumors. Still, few publications described the use of magnetic resonance in the diagnostics of lymphangiomas (11, 13). However, on the basis of available literature data its seems that magnetic resonance is the imaging method of choice. The apparent heterogeneity concerning signal enhancement (T1- and T2-dependent images) results from the variable content of water and fat in selected cysts (11). Considering the dynamic two-phase MR the cystic content is not subject to enhancement, both during the arterial and distant phase, while the internal septa are subject to enhancement in the late phase (13).

Considering the presented case the MR image of the lymphangioma was similar to that described in literature data concerning the gallbladder and other abdominal cavity organs. The CT scan showed no infiltration of surrounding structures. However, confirmation of the lymphatic nature of the cyst was not possible. Combined analysis of CT and MR suggested the possibility of a lymphangioma. Although the ultrasound examination
revealed the presence of a multilocular cyst, it was not possible to determine whether the lesion concerned the liver parenchyma or biliary ducts (initially falsely diagnosed as a lesion located in the right lobe of the liver). Final diagnosis was established on the basis of the histopathological examination – cystic hygroma.

The treatment of choice in case of lymphangioma is the complete excision of the lesion with a margin of healthy tissues, in order to prevent recurrence (1). Recently, attempts were undertaken to remove the lesions by means of laparoscopy.

CONCLUSION

The study presented a case of a 41-year old female patient diagnosed with gall-bladder and hepatoduodenal ligament lymphangioma, accidently detected during routine abdominal ultrasound as a gall-bladder and hepatoduodenal ligament cyst. Correct diagnosis was possible after combined analysis of CT and MR imaging examinations. However, the intraoperative image proved decisive. After the removal of the gall-bladder and above-mentioned lesion recurrence was not observed.

REFERENCES


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