

## Unusual co-existence of adenocarcinoma of gallbladder and retroduodenal cystic lymphangioma

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*Received 16 June 2015; accepted 14 October 2015;  
published online 24 December 2015*

### ABSTRACT

Lymphangiomas are rare benign cystic tumors. They originate from developmental failure or inflammation of lymphatics causing obstruction. More than ninety percent of lymphangioma cases are diagnosed within the first two years of life. They are mostly (95%) found in the neck and axillary regions. Intra-abdominal lymphangiomas are quite rare, and the most common locations are the retroperitoneum, mesentery, omentum and the mesocolon. Biliary tract or gallbladder cancers are often with regional lymph node metastasis but benign tumors such as lymphangioma should be kept in mind. Lymphangioma co-existing with a solid tumor might cause false over-staging and curative treatment option might be missed for some patients. A rare case of retroduodenal cystic lymphangioma co-existing with gallbladder adenocarcinoma is presented.

**Key words:** Gallbladder adenocarcinoma, retroduodenal cystic lymphangioma

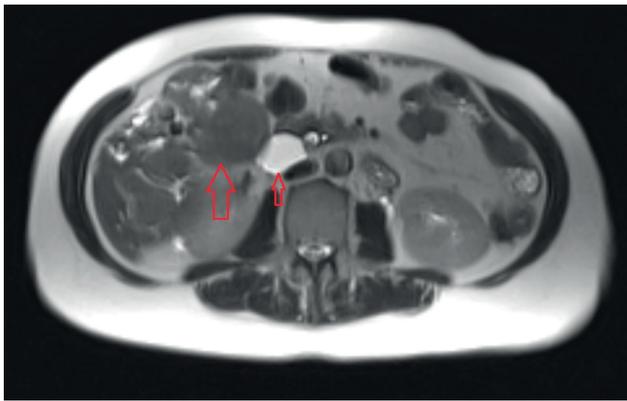
### Introduction

Lymphangiomas are benign cystic tumors and are uncommon. They originate from obstructed lymphatics caused from inflammation or developmental failure. They are mostly seen in early childhood and more than 90% of lymphangioma cases are diagnosed within the first two years of life. They are mostly found in the neck and axillary regions. Intra-abdominal lymphangiomas are quite rare, and the most common locations are retroperitoneum, small bowel mesentery, omentum and mesocolon [1-4]. Misdiagnosis of lymphangiomas in co-existing solid tumor patients as metastasis might cause over-staging. Curative surgical option might be omitted in those patients. There are only few case reports in the literature which lymphangioma co-existing with an intra-abdominal cancer [5]. Here we present a rare case of retroduodenal cystic lymphangioma co-existing with gallbladder adenocarcinoma diagnosed with postoperative pathology.

### Case history

A 64-year-old female patient presented with a 3-week history of intermittent right upper quadrant abdominal pain. The patient's prior medical history was unremarkable. A physical examination demonstrated moderate tenderness on Mc-Burney's point and a soft palpable mass in the right upper quadrant of the abdomen. Laboratory data showed leukocytosis of 15.300/mm<sup>3</sup> and an elevated gamma-glutamyl transpeptidase value of 440 U/L (normal range: 9-36 U/L). An ultrasonography of the abdomen showed a mass in the gallbladder location. Computed tomography (CT) and magnetic resonance imaging (MRI) showed 10 cm mass lesion associated with fundus of gallbladder and another 3 cm high signal-contrast repellent lesion was found in front of the vena cava (Figure 1).

The patient underwent laparotomy and liver segment 6, gallbladder, liver hilum and a cystic lesion at superior and posterior location of duodenum were resected. Histopathological examination revealed

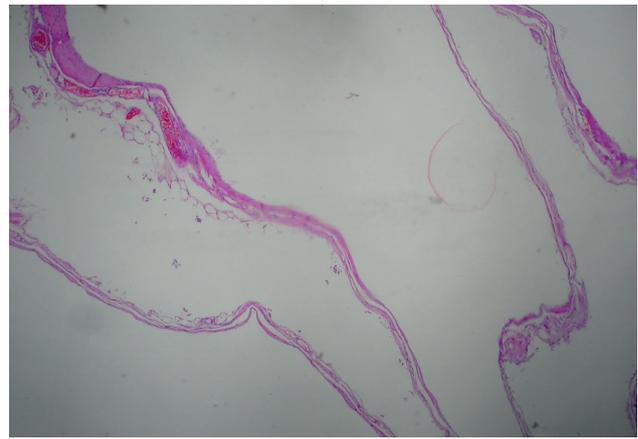


**Figure 1.** Axial T2 weighted MRI images: The mass filling the gallbladder (big arrow) and lymphangioma in front of vena cava (small arrow) in cystic density.

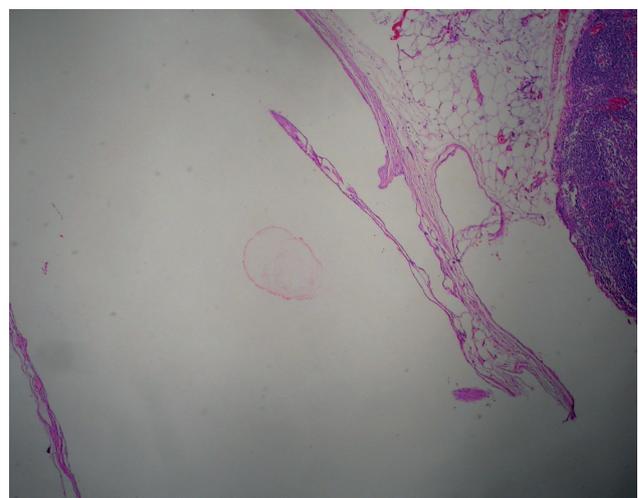
moderately differentiated adenocarcinoma of gallbladder and a metastatic lymph node at the liver hilum. Cystic lesion resected from the retro-duodenal region was histologically diagnosed as lymphangioma (Figures 2-3).

## Discussion

Etiology of lymphangiomas remain unclear and they are mostly seen in infants and children. Most commonly involved areas are neck (75%) and axilla (20%). Less than 5% of lymphangiomas are found in the intra abdominal region [1-4]. Lymphangiomas can arise from the mesentery, retroperitoneum, colon, small intestine, pancreas, omentum, mesocolon and gallbladder. Intra abdominal lymphangiomas might be asymptomatic or present with nausea, vomiting and abdominal fullness. The symptoms are due to either pressure on the adjacent structures by the enlarging mass, or to complications as hemorrhage, infection, perforation, torsion and rupture. Lymphangiomas might cause problems in the pre-surgical staging of solid tumors especially with cystic or mucinous nature. Hence, many of the previously reported cases were diagnosed incidentally during surgery. A retroperitoneal cystic lymphangioma was diagnosed co-existing with a sigmoid carcinoma in a 78-years old man in the report by Papadopoulos [6]. Patient was treated curatively and exact diagnosis of lymphangioma was given postoperatively as in our case. Ultrasonography, CT and MRI have been shown to be complimentary in the diagnosis of retroperitoneal lymphangiomas [7].



**Figure 2.** Dilated lymphatic gaps in cystic mass, hematoxyline-eosine staining.



**Figure 3.** The cystic mass and adjacent lymph node, hematoxyline-eosine staining.

## Conclusion

Differential diagnosis of periportal masses accompanying the adenocarcinoma of gallbladder is important for curative surgery. Biopsy might be helpful but it might be insufficient for exact diagnosis of cystic lesions. Periportal masses in patients with biliary tract or gallbladder cancer are often lymph node metastasis although benign tumors such as lymphangiomas should be kept in mind. Radiological imaging techniques [MRI / USG / F18-FDG positron emission tomography (F18-FDG PET CT)] may be helpful in the differential diagnosis. It should be noted that F18-FDG PET CT has a low sensitivity in mucinous tumors. Therefore, intraoperative pathological examination of periportal masses have an important place for curative treatment of biliary tract or gallbladder cancers.

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