



CYSTIC LYMPHANGIOMA OF THE GALLBLADDER: A CASE REPORT AND LITERATURE REVIEW

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<p>Article Info Received 15/02/2015 Revised 27/03/2015 Accepted 22/04/2015</p> <p>Key words: Lymphangioma; Gallbladder; Cystic.</p>	<p>ABSTRACT Lymphangioma is a benign and congenital malformation of the lymphatic system. Most are preferentially located in the head and neck region. The abdominal organs are uncommon sites of origin. The gallbladder is one of the rarest origins. Generally, intra-abdominal lymphangioma is asymptomatic and found incidentally, but in some cases, the patient complains of abdominal distension or a palpable mass. We describe the case of a 66-year-old male presented with right upper abdominal pain for 3 months long. Obsolete perforation of gallbladder and abscess were suspected, based on the findings of ultrasound and magnetic resonance imaging. A multichamber cystic tumor, 15×9×6cm in size, partly encapsulated the gallbladder, was resected <i>en bloc</i> with the gallbladder. The tumor was pathologically diagnosed as a cystic lymphangioma of the gallbladder. Since lymphangioma is benign, it could be observed with accurate diagnosis. The surgical indication would be limited to cases of symptomatic lymphangiomas.</p>
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INTRODUCTION

Lymphangiomas are benign and congenital malformations of the lymphatic system. About 95% lymphangiomas occur in the skin and the subcutaneous tissues of the head, neck, axillary, inguinal and retroperitoneal region. Patients always have no characteristic symptoms and approximately 60% of them are younger than 5 years old. Ultrasonography, computed tomography (CT) and magnetic resonance imaging (MRI) have all been used to diagnose lymphangioma. Complete surgical resection is the treatment for lymphangioma, and successful laparoscopic resection for intraabdominal lymphangioma also has been proved to be safe [1,2]. Herein we report a case of lymphangioma arising from gallbladder which is extremely rare.

Case

A 66-year-old male presented with right upper abdominal pain for 3 months long without fever and jaundice. He was otherwise in good health with no history

of ulcer and calculus. The physical findings on admission revealed tenderness at right upper abdomen, yet Murphy's symptom is negative. Laboratory data were all within normal limits. Color Doppler ultrasound showed that the gallbladder was normal in size and was encased by an irregular multicystic mass from anterior and inferior aspects, which was 85mm×69mm in size (Fig.1). No dilatation of the common bile duct and no calculus and accumulation of ascites were detected. From these findings, obsolete perforation of gallbladder, abscess were suspected.

On the next day, cholecystectomy was undertaken under general anesthesia. A multichamber cystic tumor, 15cm×9cm×6cm in size, partly encapsulated the gallbladder, was resected *en bloc* with the gallbladder. Its contents were clearing. It elongated along the anterior and posterior aspect of duodenohepatic ligament, posterior aspect and superior border of pancreas. The margin between the tumor and the gallbladder was indistinct, and



the tumor seemed to originate from the gallbladder. The gallbladder was normal in size with inspissated bile in it, but calculus was not found.

Histologically, the multicystic lump was composed of an irregularly dilated space separated by fibrous tissue. The cyst wall was lined by lymphatic

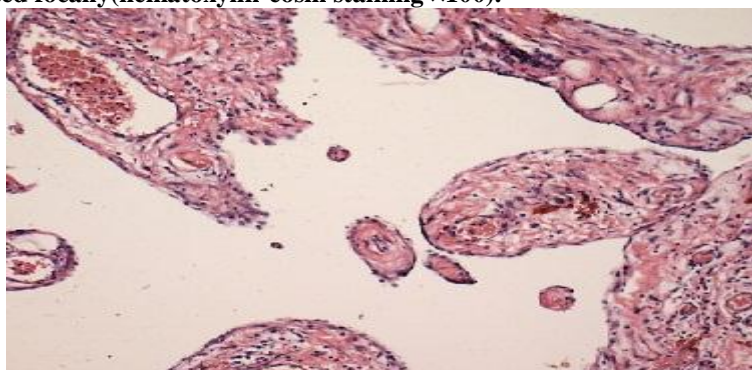
endothelial cells. Between the cyst wall, small blood vessels proliferated and lymphocytes infiltrated focally(Fig.2).

According to the evidences above, it was diagnosed as a cystic lymphangioma of the gallbladder.

Figure 1. Color Doppler ultrasound showed that the gallbladder was normal in size and was encased by a irregular multicystic mass from anterior and inferior aspects, which was 85mm×69mm in size (GB : gallbladder, L: liver, MASS:mass).



Figure 2. Histologically, the multicystic lump was composed of an irregularly dilated space separated by fibrous tissue. The cyst wall was lined by lymphatic endothelial cells. Between the cyst wall, small blood vessels proliferated and lymphocytes infiltrated focally(hematoxylin-eosin staining ×100).



DISCUSSION AND CONCLUSION

Lymphangiomas are rare, benign tumors of the lymphatic system, usually present in children aged 5 years and younger. Because they are asymptomatic until the mass enlarges to cause symptoms, most lymphangiomas are diagnosed at adulthood incidentally. A lymphangioma arising from the gallbladder represents only 0.8%-1% of all intraabdominal lymphangiomas, and only a few cases have been reported. Clinical symptoms are the pressure produced by the growing cyst, nausea, vomiting and abdominal fullness. An abdominal mass can usually be palpated. Complications are anemia, infection, obstruction of the intestine or ureter, inflammation, perforation, torsion, and rupture.

Lymphangiomas are classified histologically into three types as simple, cavernous, or cystic. Simple lymphangiomas, generally found on the skin and subcutaneous tissue of the face and neck, consist of an ill-

defined mass composed of dilated lymph channels with rich cellular connective-tissue stroma. They are connected to the adjacent lymphatic system through a moderate number of channels [3]. Cavernous lymphangiomas are generally found in children. They are located in the neck or axilla, and only rarely found in the retroperitoneum or intraabdominally. They are sponge-like compressible masses composed of communicating lymph-filled microscopic cysts separated by septa. The cystic lymphangioma, without connection with adjacent lymphatics, can be found in the neck, axilla, breast, retroperitoneum, or abdomen. Its histological characteristics include the following: flat endothelia lining the cyst, small lymphatic spaces in the cyst wall, lymphoid tissues and smooth muscle in the cyst wall [4]. The treatment of choice is complete excision which is known to be the standard treatment. If the lesion has adhered to



the surrounding organs, a total resection of the involved tissues, a partial excision may be indicated. The incomplete removal of a cystic lymphangioma may lead to its recurrence [5]. Thus, total resection of the cyst and the gallbladder was performed in our patient. Some reports suggested that gallbladder lymphangiomas were successfully treated by laparoscopic cholecystectomy. When the minimal incision is intended, downsizing the tumor volume by puncture of cyst and aspiration of cystic contents with minimum spillage is required to permit mobilization and removal of the tumor. Spillage of the cyst contents into the retroperitoneal cavity may occur during

aspiration. The SAND balloon catheter may be safe to minimize the spillage of cystic content and reduce the size under laparoscope [6].

CONFLICTS OF INTEREST

The authors do not have any conflict of interest, personal or organizational, financial or familial, real or apparent, in participating in this procurement. The author further represent that his spouse/domestic partner has no conflict of interest as defined in the Conflict of Interest Code.

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