

Inflammatory Myofibroblastic Tumor of the Gallbladder: A Review of a Potential Pitfall

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Abstract

Inflammatory myofibroblastic tumor (IMT) is a unique neoplasm that rarely spreads to other parts of the body. IMT has a wide anatomical distribution, with the mesentery, omentum, retroperitoneum, and pelvis being the most commonly affected. A careful search in the English literature resulted in finding seven cases of IMT arising from the gallbladder. Gallbladder IMT presents a clinical challenge and is preoperatively diagnosed as a malignant gallbladder tumor in most cases. Histopathological assessment is essential for correct diagnosis and management. The prognosis is favorable, with most cases showing no signs of recurrence or metastasis. Even in patients who have had surgical resection, long-term follow-up is required.

Keywords: Inflammatory myofibroblastic tumor (IMT), neoplasm, gallbladder IMT, metastasis.

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INTRODUCTION & BACKGROUND

Inflammatory myofibroblastic tumor (IMT) is a unique neoplasm that rarely metastasizes to other parts of the body. It is formed of spindle-shaped myofibroblastic and fibroblastic cells that are surrounded by inflammatory plasma cells, lymphocytes, and/or eosinophils.

IMT has a wide anatomical distribution, with the mesentery, omentum, retroperitoneum, and pelvis being the most commonly affected, followed by the lung, mediastinum, head and neck, gastrointestinal tract, and genitourinary tract, including the uterus and bladder. Somatic soft tissues, the pancreas, the liver, and the central nervous system are all unusual places. Primary gallbladder IMT is rare [1, 2].

The gene anaplastic lymphoma kinase (ALK) has recently been linked to IMT development. The ALK gene is considered to produce a fusion gene with other proteins, causing tumor cells to proliferate. Anti-ALK antibody immunostaining identifies ALK fusion gene overexpression and is thus beneficial for auxiliary IMT diagnosis [3, 4].

Malignant changes and recurrences of an inflammatory pseudotumor have been observed years after surgery, implying that long-term follow-up is

required even in patients who have had surgical resection [5].

REVIEW

This is a review for cases of primary gallbladder IMT. A careful search in the English literature resulted in finding seven cases of IMT arising from the gallbladder [Table 1].

The age of patients ranges from 35 to 66 years. Most of the patients are females (five females and two males). The tumor can be asymptomatic or can induce different signs and symptoms including abdominal pain/tenderness, nausea, vomiting, jaundice, purities and/or fever. Imaging usually reveals a tumor located in the gallbladder wall that in one case showed a polypoid protrusion into the gallbladder lumen [5].

The preoperative diagnosis of gallbladder malignancy was rendered in most cases. The differential diagnoses included inflammatory conditions like gangrenous cholecystitis, and other neoplastic processes

Three cases showed adhesion to the liver [6-8]. One case showed adhesion to the colon [9]. In those cases, the surgical resections included parts of the adhered organs.

The histopathologic assessment revealed a proliferation of spindle cells with no atypia in a background of fibrosis with inflammatory cells including plasma cells, eosinophils and histiocyte. No abnormal mitotic figures were identified [Figure 1]. The spindle cells show diffuse positivity for SMA immunohistochemical stain.

ALK immunohistochemical staining was performed on five of the seven cases. One case showed positive staining, two cases were negative and the remaining two showed equivocal staining.

The prognosis was good in all followed-up cases with no recurrence or metastases, except for one case, which came back with pancreatic head and tail tumors thirteen months later [10, 11].

Table 1: ALK: Anaplastic Lymphoma Kinase. IHC: Immunohistochemistry. CT: Computerized Tomography

First Author	Age	Sex	Clinical symptoms	Preoperative diagnosis	Surgical procedure	Gross morphology	ALK IHC Staining	Outcome
Behranwala, K.	51	F	Acute right upper abdominal pain	US suggested acute cholecystitis. At surgery, thought to be an advanced gallbladder carcinoma	An en-bloc cholecystectomy and limited transverse colectomy	The tumor measured 12 cm. Adhered to the colon	Equivocal	No local recurrence was detected at six months follow-up on CT scan
Muduly, D.	35	F	Low grade fever and dull aching, irregular pain in right hypochondrium	Gallbladder carcinoma	Radical cholecystectomy with en-bloc resection of segment IVB and V of liver and periportal, retropancreatic and retroduodenal lymphadenectomy	Mass in the fundus of gall bladder with involvement of segment V of liver.	Not performed	Disease-free at 2 years after surgery
Ozsari, I	66	M	Abdominal pain, nausea and vomiting.	Gallbladder malignancy	Cholecystectomy with a segmentary liver resection of segments 4 and 5	Mass almost completely filling the gallbladder	Not performed	Not Available
Badea, R	65	F	Abdominal pain	Gangrenous cholecystitis or Gallbladder tumor	An en-bloc cholecystectomy with a segmentary liver resection of segments 4 and 5, and lymphadenectomy of the hepatic hilum lymph nodes	Eccentrically tumoral mass of 6.5 cm with a light grayish color, bulging from the fundus of the gallbladder. The tumor infiltrated the surrounding liver parenchyma to a depth of 4 cm	Equivocal	No recurrence or metastasis in 3-month follow-up
Maruyama, Y	63	F	Abdominal fullness and pain	Not mentioned	surgical resection	tumor was detected in the gallbladder	Negative	Thirteen months following resection, tumors in the pancreatic head and tail
Yamada, T	50	M	None	Special gallbladder tumor	Gallbladder bed resection	2 cm polyp in the gallbladder fundus, and the surface was covered with a greenish black deposit	Positive	recurrence-free survival 6 years after the operation
Calvo, A	60	F	Abdominal pain, fever, pruritus and jaundice	chronic inflammatory disease or a neoplastic process	cholecystectomy	Large mass of diffuse soft tissues originating from the gallbladder, and displacing the duodenum, transverse colon and hepatic flexure	Negative	No local recurrence at three-years follow-up

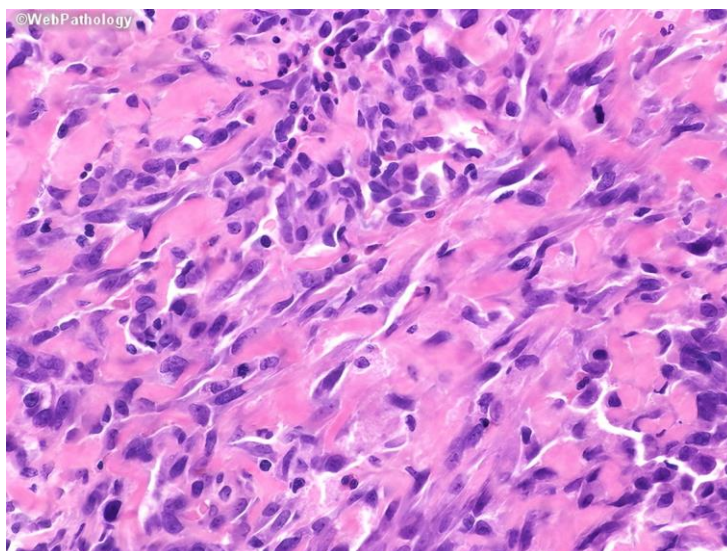


Figure 1: Myofibroblasts in fibrous background. Courtesy of Dr. Dharam Ramnani - Webpathology (with permission)

CONCLUSION

Gallbladder IMT is rarely reported. Only seven cases were reported in English literature. The tumor presents a clinical challenge and is preoperatively diagnosed as a malignant gallbladder tumor in most cases. Adhesion to adjacent organs or displacement of the intestine is seen in some cases. Histopathological assessment is essential for correct diagnosis and management. ALK immunohistochemical staining can be helpful but is not necessary, and negative or equivocal staining does not rule out the diagnosis. The prognosis is favorable, with most cases showing no signs of recurrence or metastasis. Even in patients who have had surgical resection, long-term follow-up is required.

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