



Case Report *Diagnostic Radiology*

Multimodal imaging findings of primary inflammatory myofibroblastoma of the gallbladder in adolescents

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ABSTRACT

Inflammatory myofibroblastic tumor (IMT) is a rare mesenchymal-derived neoplasm that most commonly arises in the lung. It can also occur in other parts of the body. Primary IMT of the gallbladder is exceptionally uncommon. Due to the absence of large-sample studies, the current understanding of gallbladder IMT remains largely limited to case reports. This article reports the diagnosis and treatment course of a 13-year-old male with inflammatory myofibroblastoma of the gallbladder. It specifically focuses on delineating its distinct imaging characteristics across ultrasonography, computed tomography, and magnetic resonance imaging, aiming to enhance the accuracy of pre-operative diagnosis and provide valuable insights into the literature for this rare condition.

Keywords: Gallbladder, Imaging, Inflammatory myofibroblastic tumor

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT), a rare mesenchymal lesion characterized by proliferation of fibroblasts or myofibroblasts and inflammatory cell infiltration, was first described as a granulomatous lesion by Pack and Baker in 1953.^[1] It has historically been referred to as an inflammatory pseudotumor, originally described in the lungs, but later found to occur in other tissues and organs of the body, such as the uterus and thyroid gland.^[2,3] In the abdomen, it predominantly involves the liver and gastrointestinal tract, while its occurrence in the gallbladder is extremely rare.

Given its extremely low incidence and our limited understanding of its clinical and imaging features, gallbladder IMT is often misdiagnosed as cholecystitis, gallbladder polyps, or malignancy. Previous case reports on gallbladder IMT have predominantly focused on a single type of imaging examination, whereas multimodal imaging examinations can mutually validate the diagnostic results.^[4,5] This article reports the clinical manifestations and multimodal imaging features of a case of gallbladder IMT admitted to our hospital.

CASE REPORT

A 13-year-old male, who had developed unexplained loss of appetite 6 months prior, presented to our hospital with chief complaints of nausea and vomiting, accompanied by abdominal distension. No definite signs of abdominal pain or jaundice were found during the outpatient physical examination. He also had intermittent fever, with a maximum recorded body

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temperature of 39°C. A local hospital suspected cholelithiasis complicated by chronic cholecystitis and initiated anti-inflammatory therapy, 4 months ago, which yielded no significant improvement. Consequently, he was referred to our hospital for further evaluation and management. Certain laboratory indicators were abnormal, as shown in Table 1.

Color Doppler ultrasound [Figure 1a and b] shows a cystic-solid mass in the gallbladder. Subsequently, [Figure 2a-c] CT

Table 1: Laboratory indicators.

Test item	Result	Unit	Reference range
White blood cell count	15.75	10 ⁹ /L	3.5-9.5
Hemoglobin	105	g/L	130-175
Platelet count	638	10 ⁹ /L	125-350
C-reactive protein	177.50	mg/L	0-10
Hypersensitive C-reactive protein	>5	ng/L	0-2
Tumor markers	(-)		

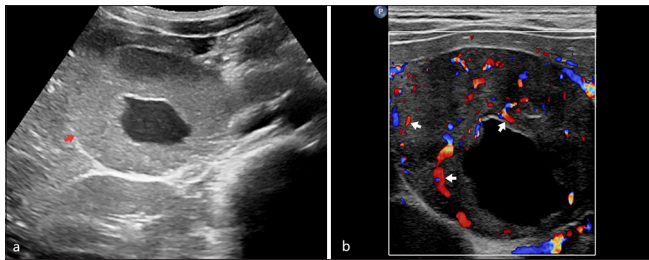


Figure 1: A 13-year-old male with a gallbladder inflammatory myofibroblastic tumor underwent routine ultrasound examination. (a) Red arrow reveals a 4.6 cm × 5.7 cm × 10.3 cm cystic-solid mass in the gallbladder with ill-defined, irregular margins, predominantly solid with a 2.4 × 4.4 cm central fluid area. (b) Color Doppler flow imaging (white arrows) demonstrates abundant vascularity within the solid component of the lesion.

and [Figure 3a-c] MRI confirmed that the gallbladder mass was predominantly solid, with a localized cystic change in the central region. [Figure 3d] shows no obvious signs of obstruction or dilatation in the intrahepatic and extrahepatic biliary systems. [Figure 3e and f] The mass showed hyperintensity on Diffusion-Weighted Imaging (DWI) and slightly hypointensity on Apparent Diffusion Coefficient (ADC) map [Figure 4a-e]. After contrast administration, the mass exhibited obvious progressive enhancement, while the central cystic change area showed no enhancement.

After consultations with the departments of general surgery, pediatrics, radiology, ultrasound, and hematology, it was decided to administer anti-inflammatory treatment and schedule a selective surgery. The patient underwent fluorescence-guided laparoscopic cholecystectomy performed by a general surgery physician. Post-operative histopathology revealed gallbladder IMT [Figure 5a and b]. Immunohistochemistry: Anaplastic Lymphoma Kinase (D5F3) (+), Brahma-Related Gene 1 (+), Integrase Interactor 1 (+), Actin (Smooth Muscle) (+), β-catenin (+), K-67 (+) (focal-10%), Actin (+), Bcl-2 (+), Cluster of Differentiation 68 (+), Cytokeratin-pan (+), Calponin (+), Epithelial Membrane Antigen (+), P53 (+). The patient underwent abdominal ultrasound re-examinations at 3 months and 1 year postoperatively, with no signs of recurrence or metastasis observed.

DISCUSSION

IMT is a rare neoplasm characterized by the proliferation of fibroblasts and myofibroblasts with inflammatory cell infiltration (composed of plasma cells, lymphocytes, and eosinophils). Gallbladder IMT is exceptionally rare, mainly reported in case studies. IMT may occur at any age, with 50-60% of cases affecting children or adolescents, and no significant sex predilection.^[6] A review of the six previously published case reports on PubMed revealed that all cases

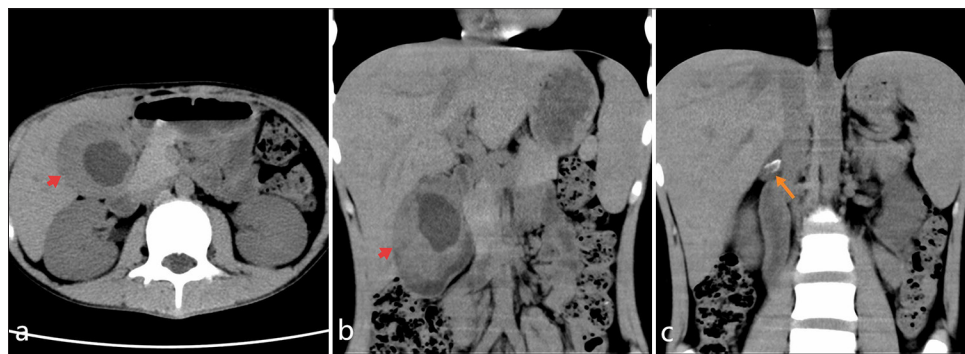


Figure 2: A 13-year-old male with a gallbladder inflammatory myofibroblastic tumor underwent a plain computed tomography (CT) scan. (a) Axial and (b) coronal non-enhanced CT (red arrows): Intraluminal gallbladder tumor containing isodense solid components (56 HU) with poorly defined margins and a central cystic component. (c) The coronal image (orange arrow) reveals an impacted gallstone in the gallbladder neck.

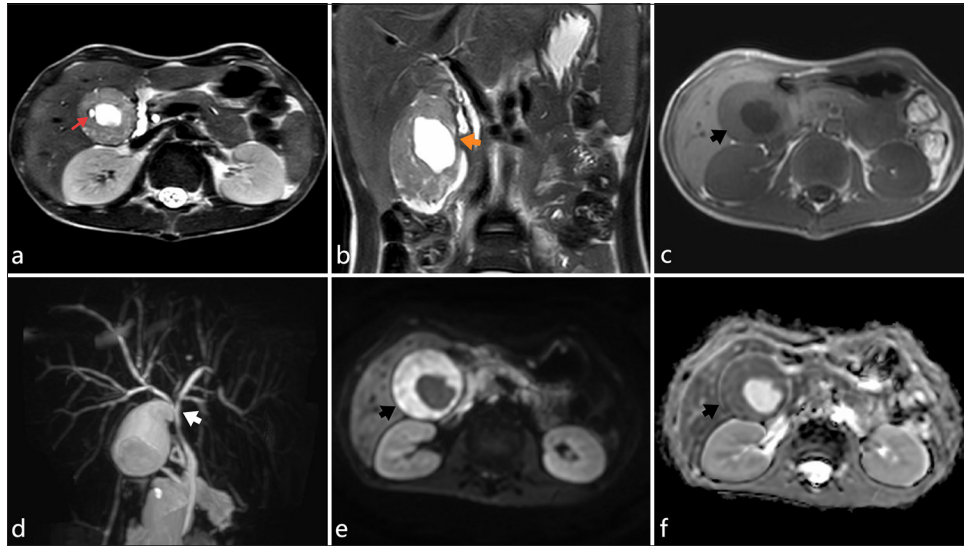


Figure 3: A 13-year-old male with a gallbladder inflammatory myofibroblastic tumor underwent a plain magnetic resonance imaging scan. (a) Axial T2WI (red arrow) shows small cystic changes within the solid component of the lesion. (b) Coronal T2-weighted imaging (orange arrow) demonstrates that the gallbladder mass arises from the mucosal wall with narrow-based attachment. (c) Black arrow T1-weighted imaging shows the solid part of the tumor isointense to hypointense, with the central cystic component appearing hypointense. (d) White arrow magnetic resonance cholangiopancreatography reveals the common bile duct and intrahepatic bile ducts, showing no significant dilation. (e) Black arrow diffusion-weighted imaging shows the solid component of the tumor as hyperintense. (f) Black arrow the apparent diffusion coefficient map shows isointense to hypointense signal in the tumor's solid component.

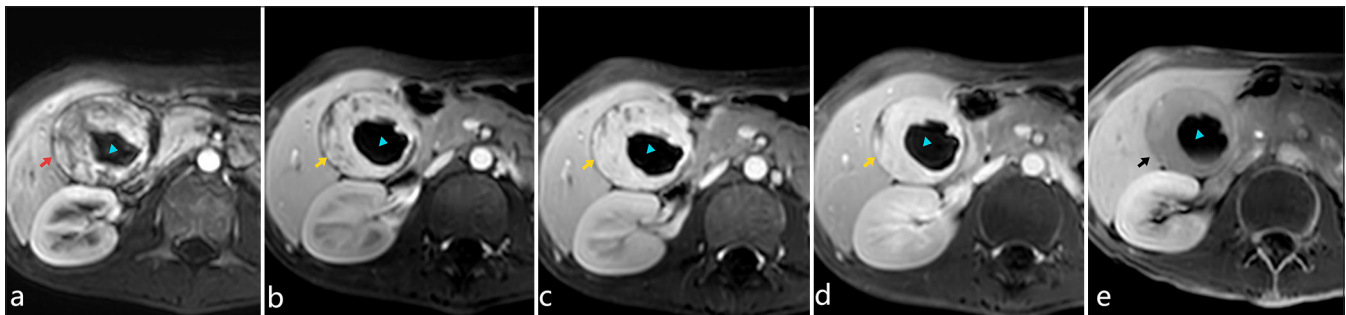


Figure 4: A 13-year-old boy with gallbladder inflammatory myofibroblastic tumor underwent contrast-enhanced MRI. ((a), red arrow) In the arterial phase of the MRI enhanced scan, the solid components of the tumor are heterogeneously enhanced. ((b-d), yellow arrows) In the portal venous phase and equilibrium phase, there is progressive and persistent enhancement. ((e), black arrow) In the delayed phase, the contrast agent gradually exits; while the cystic area (blue triangle) shows no enhancement throughout.

occurred in middle-aged and elderly populations, whereas the patient in this case is an adolescent.^[4,5,7-10] The main symptoms of this case were fever, nausea, and vomiting, which were similar to those reported in previous studies.^[7-9] In previous cases, the number of patients with abdominal pain and those without was equal, and the patient in this case showed no obvious signs of abdominal pain.

IMT typically presents as a solitary mass and requires differentiation from other common benign or malignant tumors in the corresponding location. Histopathologically,

it mainly manifests as three types:^[6] myxoid-predominant, cellular, and hypocellular. Different pathological types may lead to varying imaging manifestations. Due to the rarity of gallbladder IMT, large-sample clinical studies are lacking. The imaging features of this case include: (1) The tumor exhibits an irregular, oval-shaped appearance with localized shallow lobulation at the margin and no distinct capsule. (2) The mass is cystic-solid, with the solid component showing isodense and no obvious calcification or hemorrhage. On T1-weighted imaging, it appears

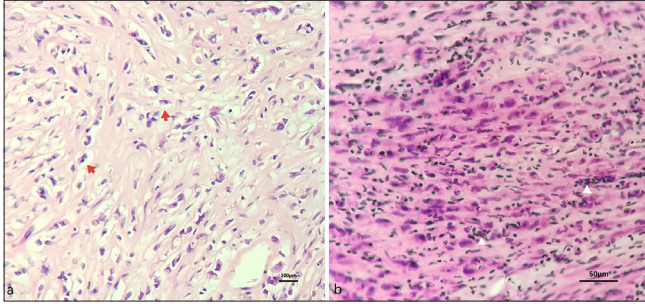


Figure 5: A 13-year-old male with a gallbladder inflammatory myofibroblastic tumor underwent a pathological examination (a) Red arrow reveals spindle cells arranged in fascicles (with moderate cellularity) within the resected tumor. The cells exhibit eosinophilic to amphophilic cytoplasm with 1-2 mitotic figures/10 high-power fields. (b) The white triangle in the stroma shows dense inflammatory cell infiltrates predominantly composed of lymphocytes and plasma cells, accompanied by scattered eosinophils and neutrophils (Hematoxylin and eosin stain a: $\times 100$ magnification, b: $\times 400$ magnification).

slightly hypointense, while on T2-7-weighted imaging, it shows mild hyperintensity with linear fine T2-hyperintense signals, likely representing mucinous components in the interstitium. The cystic portion is centrally located, relatively large, and demonstrates uniform signal intensity without layering. (3) Despite its large size, the tumor is confined to the gallbladder lumen, connected to the adjacent gallbladder wall by a narrow base [Figure 3b], with no significant inflammatory changes in the surrounding wall. (4) The solid component shows marked hyperintensity on Diffusion-Weighted Imaging (DWI), possibly due to dense arrangement of fibroblasts or myofibroblasts. (5) Post-contrast imaging revealed progressive enhancement of the solid portion, which may be correlated with fibroblast and myofibroblast proliferation, leading to contrast retention in the perivascular space. (6) No signs of invasion of the adjacent liver are observed, and no enlarged lymph nodes are detected in the hepatic hilum or retroperitoneum.

An analysis of all cases reveals that all presented with gallbladder wall thickening or space-occupying lesions. Most of these lesions were cystic-solid, showing mild-to-moderate enhancement after contrast administration. Among them, one case exhibited a “fast-in and fast-out” enhancement pattern,^[5] which was completely opposite to the progressive enhancement observed in the current case. This difference is thought to be potentially associated with the varying proportions of components within the mass. In addition, some of the cases were accompanied by gallstones,^[5,7,9] which is consistent with the present case. None of the cases developed recurrence or metastasis, which suggests a favorable tumor prognosis.

From an imaging perspective, gallbladder IMT requires differentiation from the following conditions: (1) Gallbladder carcinoma: It typically manifests as an irregular-shaped mass in the gallbladder region. The mass often broadly invades the entire gallbladder wall, causing wall thickening and architectural distortion. It frequently extends into adjacent tissues (e.g., liver and porta hepatis vessels), resulting in pericholecystic fluid collections and intrahepatic biliary duct dilation. (2) Gallbladder adenoma: It is a benign tumor originating from the gallbladder glandular epithelium and is classified as a premalignant lesion. It typically presents as a solitary, well-defined nodule protruding into the gallbladder lumen, with preserved wall architecture and distinct layering. On contrast-enhanced imaging, it often demonstrates homogeneous and marked enhancement during the arterial phase. (3) Xanthogranulomatous cholecystitis: It commonly features gallbladder enlargement with diffuse wall thickening, containing non-enhancing hypodense nodules. Due to the preservation of the mucosal line, it may display a characteristic “sandwich sign.”

Treatment options for IMT are diverse, with surgical resection serving as the primary treatment approach.^[6] Most patients achieve a favorable prognosis following curative treatment, while a minority experience post-operative recurrence, necessitating long-term clinical surveillance.

CONCLUSION

Given the low incidence and high misdiagnosis rate of gallbladder IMT, systematic analyses of its diagnosis, treatment, and pathology require larger-scale cohort studies. Due to the tumor’s potential for malignant transformation, recurrence, or metastasis, structured long-term clinical surveillance is essential following treatment.

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