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Case Report and Review of the Literature

Gallstone Ectopia: A Rare Clinical Entity

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ARTICLE INFO

Article history:

Received: 28 November, 2022

Accepted: 27 December, 2022

Published: 24 February, 2023

Keywords:

Gallstone ectopia

cholecystectomy

lung

ABSTRACT

Gallstone ectopia to the lung is a rare clinical entity. Our case is a 69-year-old man with a history of cholecystectomy complicated by intra-abdominal abscesses and retained gallstone remnants requiring laparoscopic extraction. He presented with hemoptysis and imaging showed multiple calcified lung nodules. There was a concern for gallstone ectopia in the lungs. He underwent a right middle lobectomy via thoracotomy with gallstone extraction. The patient did well postoperatively and was discharged home on postoperative day 4. This case report briefly reviews the literature to determine the timeline for presentation for similar patients, which may be of clinical utility to thoracic surgeons.

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Introduction

Gallstone ectopia to the lung is a rare entity documented in less than 20 case reports [1, 2]. Classically, patients present with malaise or hemoptysis and are found to have an abscess or mass in the right lower lobe. Diagnosis is typically made after surgical resection when pathology reveals gallstones, and surgical history involves a history of cholecystectomy or gallbladder perforation.

We report the case of a 69-year-old male who presented with hemoptysis and had a history of perihepatic abscesses from retained gallstones. Despite subtle radiographic findings, given his history, we had a high index of suspicion for gallstone ectopia. Thoracic exploration confirmed the diagnosis. A literature review is included to demonstrate the variety of possible presentations for this rare clinical entity. The review should encourage clinicians to have a higher index of suspicion for this diagnosis in patients with the appropriate surgical history.

Case Report

Our patient is a 69-year-old man with a history of hypertension, benign prostatic hyperplasia, and metastatic renal cell carcinoma (RCC) presenting with small-volume hemoptysis. He had undergone multiple abdominal operations, including left adrenalectomy, cholecystectomy for symptomatic cholecystitis, and right adrenalectomy and nephrectomy. Six months after his last operation, he was readmitted for malaise and found to have perihepatic abscesses concerning RCC metastasis. On laparoscopic exploration, retained gallstone remnants were identified and extracted. Intraoperative cultures were positive for *Actinomyces*. Six months later, he developed a sinus tract, and imaging again demonstrated additional intra-abdominal abscesses. A mini laparotomy was required to drain an abscess on the right abdominal wall at the costal margin and in the midepigastrium.

A year later, he presented with hemoptysis. A computed tomography scan of the chest was obtained (Figure 1). Multiple calcified lung nodules were noted in the right middle and lower lobes. This was initially

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concerning for RCC metastasis, but given his history, there was also a concern for gallstone ectopia in the lungs. He was referred to thoracic surgery. Intra-operatively, the middle and lower lobes were densely adherent to the diaphragm and required extensive decortication. He underwent right video-assisted thoracoscopic surgery (VATS) converted to open thoracotomy and total pulmonary decortication. A fistulous

connection to the middle lobe containing three gallstones was identified (Figure 2). A right middle lobectomy with gallstone extraction was performed. Intraoperative cultures grew *Enterococcus faecalis*. Final surgical pathology showed parenchymal fibrosis surrounding a cavity containing gallstones. He was discharged on postoperative day four and is doing well postoperatively.

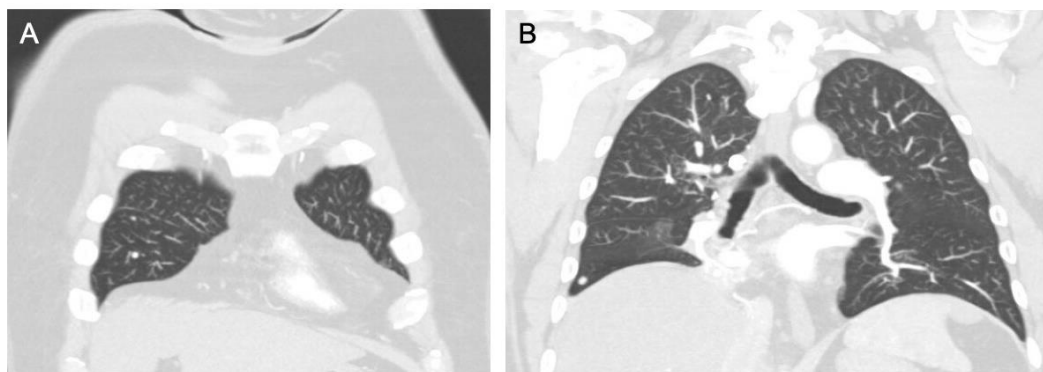


Figure 1: Computed tomography scan of chest with intravenous contrast showing A) two hyperenhancing nodules in the right middle lobe of the lung and B) one hyperenhancing nodule in the right lower lobe of the lung.



Figure 2: Gross pathology specimen with 3 gallstones extracted from lung.

Discussion

This is a rare case of gallstone ectopia causing hemoptysis that required lobectomy. Despite subtle radiographic findings, surgical treatment was pursued quickly due to a high index of suspicion, given the patient's surgical history.

Gallstone ectopia to the lung is documented in less than 20 case reports [1, 2]. Often, pre-operative imaging reveals an abscess or mass in the right lower lobe of the lung (Table 1). The differential for these findings is broad and includes infection, malignancy, or benign tumors. Radiographic findings are insufficient for a definitive diagnosis. Gallstone ectopia is usually diagnosed with final pathology, and surgical history reveals a history of cholecystectomy or gallbladder perforation.

Table 1: Brief literature review of gallstone ectopia.

Investigator	Presenting Symptoms	Prior Cholecystectomy or Gallbladder Procedure?	Time from Surgery to Presentation	Imaging Findings	Treatment
Fontaine 2006 [1]	Hemoptysis	Laparoscopic Cholecystectomy	34 months	7 cm pulmonary infiltrate with calcification in RLL	Right thoracotomy with 2 cm diaphragmatic resection IV antibiotics
Zhang 2014 [2]	Hemoptysis	Intrahepatic gallstone removal surgery with partial hepatectomy	5 months	4.4 cm solid mass in the RLL with calcifications	RLL wedge resection

Binmahfouz 2016 [3]	Anorexia and weight loss	Complicated open cholecystectomy with gallbladder spillage	3 years	2.8 cm PET avid mass with central calcification in RLL	RLL wedge resection
Quail 2014 [4]	Hemoptysis	Laparoscopic Cholecystectomy	5 years	3 hypodense foci in RLL with peridiaphragmatic inflammation	RLL decortication and wedge resection
Willekes 2009 [5]	Fever, right pleuritic chest pain, right upper quadrant pain	Laparoscopic Cholecystectomy	17 months	RLL fluid collection	RLL decortication
Ianniti 2006 [6]	Generalized malaise	Laparoscopic Cholecystectomy	4 years	Trapped RLL with effusion 3 cm right subphrenic abscess	RLL decortication Abdominal abscess drainage IV antibiotics
Houghton 2005 [7]	Hemoptysis	Laparoscopic Cholecystectomy	3.5 years	3 cm mass in the RLL containing calcifications, hilar lymphadenopathy	Right thoracotomy with wedge resection of RLL mass
Werber 2001 [8]	Massive hemoptysis	Laparoscopic Cholecystectomy	1 year	4 cm RLL density with right hemidiaphragm	Right thoracotomy with RLL wedge resection Oral antibiotics
Barnard 1995 [9]	Right-sided pleuritic pain	Laparoscopic Cholecystectomy	6 months	Right subphrenic abscess with calcified bodies	Right thoracotomy with right middle lobectomy
Lee 1993 [10] *2 pts	Pt 1 and 2: Cholelithoptysis, Pt 1: Malaise	Laparoscopic Cholecystectomy	Pt 1: 4 months Pt 2: 1 year	Pt 1: Right middle lobe atelectasis Pt 2: RLL consolidation with 3-5 mm calcified nodules	Pt 1: Repeat ERCP, abdominal wound exploration with abx Pt 2: Repeat ERCP and laparotomy

PET: Positron Emission Tomography; RLL: Right Lower Lobe; IV: Intravenous; Pt: Patient; ERCP: Endoscopic Retrograde Cholangiopancreatography.

The average time to presentation from initial gallbladder intervention was 2.1 years in our literature review (Table 1), likely due to these factors: gallstone ectopia is uncommon, has initially subtle radiographic findings, and tends to lead to more indolent infections [1-10]. Seven cases with a non-operative resolution of gallstone ectopia were excluded from this analysis [9, 11-15]. By the time thoracic resection was performed, most cases were associated with hemoptysis and marked imaging findings such as a 3-7 cm infiltrate (Table 1) [1-3, 6-8].

The pathophysiologic mechanism of this disease is gallbladder spillage at the time of cholecystectomy and subsequent diaphragmatic erosion or weakening, resulting in gallstone translocation to the lung (Figure 3). Approximately 700,000 cholecystectomies are performed annually in the United States [16]. Gallbladder spillage occurs in 6-40% of laparoscopic, cholecystectomies with one-third of these incidents resulting in retained stones that were not retrieved [16, 17]. With this number of cases annually, there are many opportunities for future gallstone ectopia.

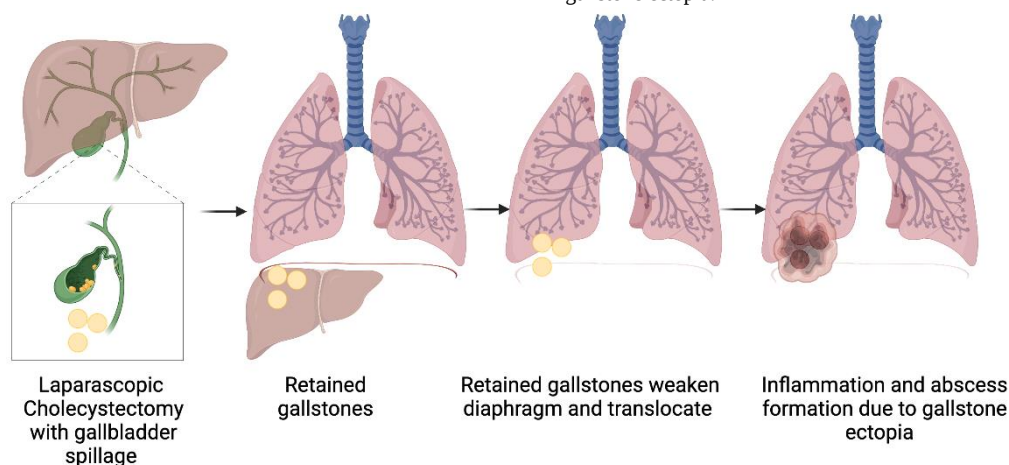


Figure 3: Pathophysiology of gallstone ectopia.

In the appropriate clinical context, a high index of suspicion should be maintained for gallstone ectopia. This should include all patients with a history of cholecystectomy who present with hemoptysis or malaise and have what appears to be a calcified lung nodule on imaging. In our case, a high index of suspicion led to early therapeutic intervention with lobectomy prior to the development of a large pulmonary abscess or pleural effusion, and the patient had an uneventful postoperative course and rapid recovery.

Acknowledgement

The authors would like to acknowledge the support of the University of North Carolina School of Medicine and the Department of Surgery for this project. Figure 3 was created with BioRender.com.

Conflicts of Interest

None.

Funding

None.

Abbreviation

ERCP: Endoscopic Retrograde Cholangiopancreatography

IV: Intravenous

PET: Positron Emission Tomography

Pt: Patient

RCC: Renal Cell Carcinoma

RLL: Right Lower Lobe

VATS: Video-Assisted Thoracoscopic Surgery

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