

# Hepatic Actinomycosis in a Patient With Retained Common Bile Duct Stent

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## ABSTRACT

Primary hepatic actinomycosis is rare, with less than 100 cases reported in English literature. Most of these cases are cryptogenic. We describe a 35-year-old woman who presented with a retained common bile duct stent for 6 years and found to have a hepatic mass with altered perfusion and enhancement, and minimal degree of washout on enhanced cross-sectional imaging. Fine-needle aspiration revealed presence of filamentous bacteria morphologically consistent with *Actinomyces* species. This report is a demonstration of a rare instance in which a retained biliary stent led to primary hepatic actinomycosis.

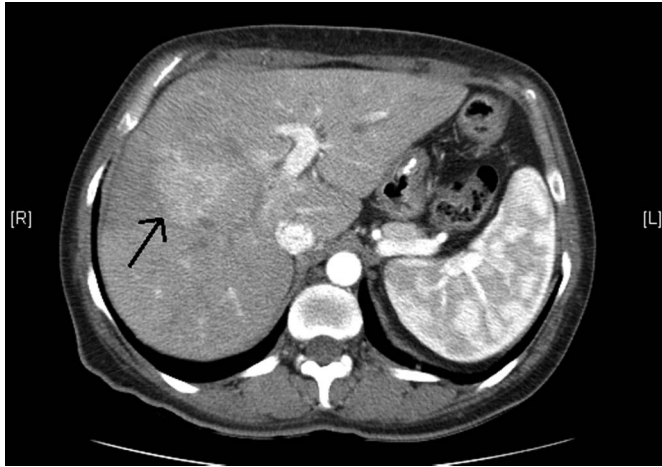
## INTRODUCTION

Actinomycosis is rare, with most patients presenting with chronic, cervicofacial soft tissue abscesses. Abdominal actinomycosis represents a minority of cases, with hepatic actinomycosis representing only one-fourth of intra-abdominal occurrences.<sup>1</sup> When hepatic actinomycosis does occur, it is usually secondary to another intra-abdominal source.<sup>2</sup> There are no previous cases reporting retained biliary stents causing primary hepatic actinomycosis. We describe a case of hepatic actinomycosis presenting as a liver mass lesion in a patient with retained biliary stent.

## CASE REPORT

A 35-year-old woman presented with 6 weeks of severe upper abdominal pain, anorexia, subjective fevers, and constipation. Her history is significant for a previous cholecystectomy, complicated by a bile leak. Common bile duct stent placement was performed through endoscopic retrograde cholangiopancreatography 6 years ago and the stent had never been removed. Other previous surgeries included a cesarean section, hysterectomy, and gastric sleeve. Initial laboratories revealed white blood cell count 12,000 cells/mm<sup>3</sup>, platelets 549,000 cells/mm<sup>3</sup>, erythrocyte sedimentation rate 56 mm/hr, C-reactive protein 58 mg/L, albumin 4.0 g/dL, gamma glutamyl transpeptidase 73 U/L, total bilirubin 0.6 mg/dL, alanine aminotransferase 14 U/L, aspartate aminotransferase 16 U/L, and total alkaline phosphatase 159 U/L. Abdominal computed tomography scan revealed that the stent had migrated upstream to the superior aspect of the common bile duct. Also seen was a hepatic lesion with heterogeneous enhancement measuring 5.6 × 4.2 cm. The area of heterogeneous enhancement showed subsequent degree of Isovue-370 contrast washout in the postarterial phase (Figure 1). The anterior branch of the right portal vein seemed to be involved.

Endoscopic retrograde cholangiopancreatography was performed for stent removal. A stent with occlusion was removed from the common bile duct. Mild biliary narrowing was found in the lower third of the common bile duct. Further workup of the hepatic mass was pursued. Abdominal ultrasound showed a focal area of marked attenuation of the bile ducts with mild peripheral ductal dilation, concerning for cholangiocarcinoma. Tumor markers including alpha-fetoprotein, carcinoembryonic antigen, CA 19-9 antigen, and



**Figure 1.** Segment 5 of the liver showing area of heterogeneous ill-defined enhancement measuring roughly  $4.2 \times 5.6$  cm is seen (arrow) exhibiting subsequent degree of washout. Distal to this region is altered perfusion of the liver parenchyma.

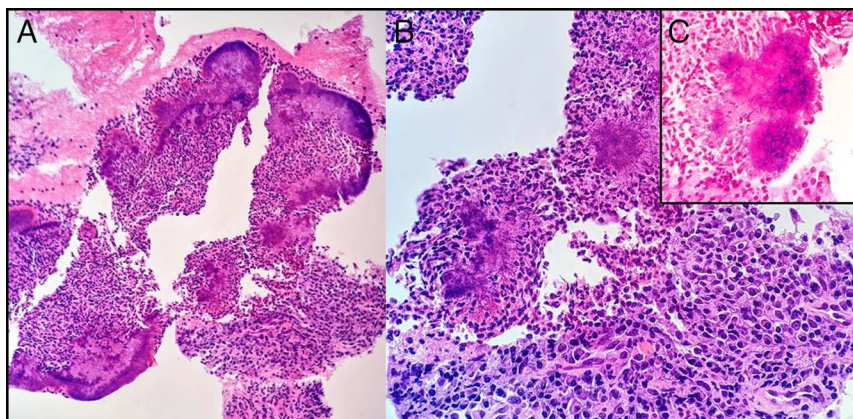
CA 125 antigen were within normal limits. Ultrasound-guided fine-needle aspiration was performed. The aspirate was negative for malignancy and revealed the presence of Gram-positive filamentous bacteria morphologically consistent with *Actinomyces* species, adjacent to hepatic parenchyma with mixed acute and chronic inflammation and granulation tissue (Figure 2).

Infectious Disease experts were consulted for the management of hepatic actinomycosis. Further workup including serum histoplasma, serum cryptococcal antigen, aspergillus, and beta-D-glucan were negative. Human immunodeficiency virus screen was negative. She was placed on ampicillin/sulbactam to complete 6 weeks of intravenous antibiotic therapy. Repeat cross-sectional imaging with Eovist-enhanced MRI showed a dramatic decrease in size of the lesion seen at presentation, measuring  $3.4 \times 1.6$  cm (Figure 3). She is currently being followed in the Infectious Disease clinic to continue with her antibiotic course.

## DISCUSSION

Actinomycosis infectious is a rare, chronic infection caused by Gram-positive bacilli of the *Actinomyces* genus. Cervicofacial actinomycosis is the most frequent presentation, as *Actinomyces* are part of the commensal flora of the oropharynx and gastrointestinal tract. Abdominal actinomycosis is less frequent, comprising about 20% of cases.<sup>1</sup> Most of these cases occur in the appendix or ileocecal region, with hepatic infection occurring in ~5% of this already rare infectious process.<sup>1</sup> Most hepatic actinomycosis cases arise from other intra-abdominal infections resulting from loss of integrity of the gastrointestinal mucosa, previous abdominal surgeries, foreign bodies, or immunosuppression.<sup>2</sup> In this case, the patient had a sphincterotomy with a retained common bile duct stent, which most likely caused a breach in the gastrointestinal integrity and the presence of foreign body in the biliary tree, serving as the nidus of infection. In addition, this patient had a previous cholecystectomy complicated by a bile leak, which could further be an additional risk factor for the development of abdominal actinomycosis in this particular scenario.

Primary hepatic actinomycosis is rare, with less than 100 reported cases in English literature from the 1960s to 2015. Many cases masquerade as malignancies. Reports have demonstrated that these infections may appear as solitary, malignant processes in almost half of the cases.<sup>3</sup> This case was not an exception, as malignancy was at the top of the differentials until fine-needle aspiration was completed. What further complicates diagnosis is the wide variety of symptoms found on presentation. In this case, the patient had severe upper abdominal pain, anorexia, and subjective fevers. However, not all patients with hepatic actinomycosis have abdominal pain or fever, and some demonstrate constitutional symptoms such as night sweats and weight loss.<sup>1,4,5</sup> Her bilirubin level was normal, which was thought to be due to adequate biliary drainage despite partial occlusion of the biliary stent, and the location of the hepatic lesion in proximity to the common bile duct.



**Figure 2.** (A) Hematoxylin and eosin stain (200 $\times$ ) of liver biopsy showing suppurative and granulomatous inflammation with scattered “Splendore–Hoeppli” phenomenon (basophilic centers with eosinophilic periphery). (B) Hematoxylin and eosin stain (400 $\times$ ) of liver biopsy showing thin, filamentous organisms abutting reactive tissue. (C) Gram stain (600 $\times$ ) showing Gram-positive filamentous bacteria.



**Figure 3.** Magnetic resonance imaging of hepatic lesion post antibiotics demonstrating decrease in size 8 weeks after treatment.

Definitive diagnosis of hepatic actinomycosis depends on histochemical and microscopic evaluation of tissue specimens, as imaging studies are not specific. The majority of computed tomography images demonstrate a hypoattenuated lesion, which cannot rule out hepatic malignancy.<sup>2</sup> A biopsy of the lesion must be performed for definitive diagnosis. Pathologic examination may reveal sulfur granules, and filamentous Gram-positive bacteria can be seen on Gram stain.<sup>2</sup>

Treatment options involve antibiotics, surgical resection, and lesion drainage percutaneously. These interventions can be done separately or in combination.<sup>1</sup> Guidelines are lacking regarding when singular or combination therapy is warranted; however, good prognosis has been found in both scenarios.<sup>1</sup> In this case, the patient responded well to antibiotic therapy alone. Penicillin derivatives are recommended. Patients with actinomycosis infections require a 6- to 12-month course of antibiotic therapy to adequately penetrate the infected tissues if not surgically resected.<sup>6</sup> Refractory cases may require surgical resection.<sup>7</sup>

Studies investigating the complications of forgotten long-term biliary stents have been performed. The most common complication seems to be acute cholangitis associated with common bile duct stones.<sup>8</sup> Although there have been case reports documenting hepatic actinomycosis secondary to intra-abdominal actinomycosis in patients with foreign bodies (ie, intrauterine contraceptive devices), there is scarce of data in the literature reporting retained biliary stents as a leading cause of primary hepatic actinomycosis.<sup>2</sup> Common bile duct actinomycosis was previously reported in a case of retained common bile duct stent.<sup>9</sup> However, in that case, the patient also had an intrauterine device in place, which could have also served as a nidus for infection.<sup>10</sup>

This case demonstrates yet another complication of retained bile duct stents in immunocompetent individuals, illustrating the importance of having ancillary staff in established advanced endoscopy programs for follow-up on cases after placement of biliary and/or pancreatic duct stents. It also provides an example of the importance of keeping primary hepatic actinomycosis in the differential in similar scenarios, as those lesions can be easily mistaken as neoplastic processes.

## DISCLOSURES

Author contributions: A. Grossen interpreted data, wrote and revised the manuscript. M. Magguilli provided the pathology images and revised the manuscript. TC Thai provided the radiology images, edited and revised the manuscript. G. Salem revised and approved the final version, and is the article guarantor.

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Informed consent was obtained for this case report.

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## REFERENCES

1. Avila F, Santos V, Massinha P, et al. Hepatic actinomycosis. *GE Port J Gastroenterol.* 2015;22(1):19–23.
2. Yang XX, Lin JM, Xu KJ, et al. Hepatic actinomycosis: Report of one case and analysis of 32 previously reported cases. *World J Gastroenterol.* 2014; 20(43):16372–6.
3. Kanellopaoulou T, Alexopoulou A, Tanouli MI, et al. Primary hepatic actinomycosis. *Am J Med Sci.* 2010;339(4):362–5.
4. Lall T, Shehab TM, Valenstein P. Isolated hepatic actinomycosis: A case report. *J Med Case Rep.* 2010;4:45.
5. Tiftikci A, Vardareli EN, Kaban K, Peker O, Akansel S, Tozun N. Actinomycotic hepatic abscess. *Hepatol Int.* 2008;2(1):133–5.
6. Valour F, Sénéchal A, Dupieux C, et al. Actinomycosis: Etiology, clinical features, diagnosis, treatment, and management. *Infect Drug Resist.* 2014;7: 183–97.
7. Wong VK, Turmezei TD, Weston VC. Actinomycosis. *BMJ.* 2011;343: d6099.
8. Sohn SH, Park JH, Kim KH, Kim TN. Complications and management of forgotten long-term biliary stents. *World J Gastroenterol.* 2017;23(4):622–8.
9. Thadani A, Chick J, Gollogly H, Galandauer I, Pais S. Actinomycosis of the common bile duct diagnosed by endoscopic ultrasound fine-needle aspiration. *Gastroenterol Hepatol (N Y).* 2010;6(11):725–7.
10. Yang SS, Im YC. Severe abdominopelvic actinomycosis with colon perforation and hepatic involvement mimicking advanced sigmoid colon cancer with hepatic metastasis: A case study. *BMC Surg.* 2018;18(1):51.

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